

**ENGAGEMENT OF INTEREST-HOLDERS IN EVIDENCE SYNTHESIS AND  
HEALTH GUIDELINE DEVELOPMENT**

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## PREFACE

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The research described in Chapter 5 (article 3) received ethical approval from the Bruyere Research Institute (#M16-21-012), Ottawa, Canada. Ethical approval was not required for other chapters included in this thesis.

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## ABSTRACT

Evidence syntheses and guidelines are crucial in evidence ecosystems. These ecosystems ensure that relevant and timely evidence is fit-for-purpose and utilized for health decision-making by interest-holders such as patients, providers and policymakers. Presently, there is a lack of guidance on how to meaningfully engage interest-holders in evidence synthesis and guideline development. The MuSE Consortium aims to address this gap by developing equity-oriented guidance for engagement in these processes. This thesis, part of the MuSE Consortium's research program, addresses knowledge gaps related to equity, barriers and facilitators, and principles and strategies for engagement of interest-holders in evidence synthesis and guideline development. We conducted a scoping review of evidence synthesis literature, identifying considerations for engagement across equity characteristics and summarizing principles to support equitable engagement. Additionally, our qualitative evidence synthesis examined barriers and facilitators to engagement in health guideline development, mapping findings to the GIN-McMaster Checklist for Guideline Development. We also interviewed 43 interest-holders to identify principles and strategies to promote their engagement in the guideline development process.

Our scoping review revealed that engagement in evidence synthesis is concentrated in high-income English-speaking countries, with technology access issues leading to resource bias and exclusion of people from lower socio-economic positions. We identified four principles for equitable engagement: addressing power dynamics, ensuring accessibility, promoting diversity and inclusion, and ethical considerations. Our qualitative evidence synthesis identified 24 barriers and 28 facilitators for engagement in health guideline development. Key facilitators included implementing an organizational engagement model, recruiting knowledgeable and diverse interest-holders, defining clear roles and responsibilities, providing training and support, and addressing power dynamics. Interviews with stakeholders underscored the importance of horizontal power structures and shared decision-making. Participants emphasized the need for epistemic justice, promoting fairness and equality in knowledge production. Strategies included engaging interest-holders in governance, setting clear expectations and timelines, fair compensation, and meaningful engagement throughout the process. These findings are widely applicable to any organization or government producing evidence syntheses, health recommendations, or guidelines. Equitable engagement ensures that all voices are heard and valued in decision-making processes, addressing power imbalances and social inequities, and leading to more effective health interventions and outcomes.



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## **LIST OF ABBREVIATIONS**

|               |                                                                                                                                                                               |
|---------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| GIN           | Guidelines International Network                                                                                                                                              |
| GDC           | GIN-McMaster Guideline Development Checklist                                                                                                                                  |
| MuSE          | A global consortium of over 160 individuals interested in engaged research methods, previously the “Multi-Stakeholder Engagement” (MuSE) Consortium                           |
| PRISMA        | Preferred Reporting Items for Systematic reviews and Meta-Analyses                                                                                                            |
| PROGRESS-Plus | Place of residence, race/ethnicity/culture/language, occupation, gender or sex, religion, education, socio-economic status, social capital, age (plus) and disability (plus). |

## CHAPTER 1: INTRODUCTION

### 1.1 Rationale

Evidence ecosystems are dynamic systems that facilitate the continuous production, synthesis, and application of research evidence to inform health decision-making (Gough et al., 2019; Gough et al., 2020; Elliott et al., 2014). These ecosystems integrate multiple components including primary research, evidence syntheses, clinical practice guidelines, and decision support tools to create a seamless flow of evidence from generation to real-world implementation. The goal is to ensure that high-quality, relevant, and timely evidence is available and utilized across all levels of healthcare, from individual patient care to public health policy.

The concept of evidence ecosystems emphasizes the interconnectedness of various interest-holders including researchers, healthcare providers, policymakers, and patients, in the process of evidence production and utilization. By fostering engagement among these groups, evidence ecosystems aim to improve the efficiency and impact of research on healthcare outcomes. They also focus on addressing barriers to evidence uptake and enhance the dissemination and implementation of evidence in clinical practice.

Presently, there is a lack of guidance on how to meaningfully and equitably engage a broad range of interest-holders in two components of evidence ecosystems: evidence synthesis and guideline development. The MuSE Consortium (previously, the Multi-Stakeholder Engagement Consortium) aims to address this gap by developing evidence-based and equity-oriented guidance documents for evidence synthesis researchers and guideline developers on how to engage 11 different groups of interest-holders in these processes. The articles included in this thesis fall within the research program of the MuSE Consortium and address knowledge gaps related to equity, barriers and facilitators, and principles and strategies for engagement of interest-holders in evidence synthesis and guideline development.

## 1.2 Objectives

This thesis examines approaches for interest-holder engagement in evidence synthesis and health guideline development. Its goal is to inform the guidance developed by the MuSE Consortium, which aims to provide equity-oriented guidance for use by evidence synthesis researchers and guideline developers for the engagement of 11 different groups of interest-holders. Specifically, this thesis addresses the following objectives:

1. To critically examine the definitions of “interest-holder” and “engagement” in the evidence synthesis literature;
2. To identify considerations to promote equitable engagement in evidence synthesis and support the engagement of interest-holders across equity characteristics (defined by PROGRESS-Plus);
3. To synthesize the reported evidence on barriers and facilitators to engagement for interest-holders in health guideline development;
4. To explore interest-holders’ perspectives on principles and strategies to promote their engagement in the development of health guidelines.

## 1.3 Thesis organization

This thesis is an article-based thesis and is organized as follows:

*Chapter 2* provides background information on the following key concepts: (1) Situating this thesis within the research program of the MuSE Consortium, (2) definition of interest-holders in health research, (3) the rationale and existing frameworks for engagement in health research, (4) evidence ecosystems, and (5) current knowledge gaps for engaging interest-holders in evidence synthesis and health guidelines.

*Chapter 3* addresses the first two research objectives stated above. The article is a scoping review which examines definitions for “interest-holder” and “engagement” in the evidence synthesis literature. Further, it identifies considerations for engaging interest-holders across the equity characteristics defined by PROGRESS-Plus: (O’Neill et al., 2014): Place of residence,

race/ethnicity/culture/language, occupation, gender or sex, religion, education, socio-economic status, social capital, age (plus) and disability (plus). The paper also summarizes principles to support equitable engagement in evidence synthesis research. This article has been prepared for submission to a Special Collection of *Cochrane Evidence Synthesis and Methods* (anticipated Fall 2024).

*Chapter 4* addresses the third research objective. The article is a qualitative evidence synthesis which summarizes barriers and facilitators to engaging interest-holders in health guideline development. This article specifically examines engagement across the topics of the Guidelines International Network (GIN)-McMaster Guideline Development Checklist (GDC). This article has been submitted for publication to *Campbell Systematic Reviews* (submitted February 2024).

*Chapter 5* addresses the fourth research objective. It presents a qualitative study based on 43 interviews conducted among interest-holders about their engagement in health guideline development. The article explores the principles and strategies that interest-holders believe are important to promote their engagement in the guideline development process. This article has been prepared for publication in the *BMC Medical Research Methodology* collection “Inclusive methodological awareness for equity and diversity”.

*Chapter 6* is an overall discussion that brings together the results of the three papers. It concludes the thesis with a discussion of the overall practice and research implications of the body of research and provides recommendations for future research.

## **1.4 References**

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O'Neill, J., Tabish, H., Welch, V., Petticrew, M., Pottie, K., Clarke, M., ... & Tugwell, P. (2014). Applying an equity lens to interventions: using PROGRESS ensures consideration of socially stratifying factors to illuminate inequities in health. *Journal of clinical epidemiology*, 67(1), 56-64.

## **CHAPTER 2: BACKGROUND**

### **Chapter overview**

As introduced in Chapter 1, this thesis focuses on interest-holder engagement in evidence synthesis and health guideline development. Chapter 2 provides background information on the following key concepts: (1) Situating this thesis within the research program of the MuSE Consortium, (2) definition of interest-holders in health research, (3) the rationale and existing frameworks for engagement in health research, (4) evidence ecosystems, and (5) current knowledge gaps for engaging interest-holders in evidence synthesis and health guidelines.

### **2.1 The research program of the MuSE Consortium**

This thesis falls within the research program of the MuSE Consortium. The MuSE Consortium (previously, the Multi-Stakeholder Engagement Consortium) is a group of over 160 individuals from over 30 countries with an interest and expertise in engaged health research. The MuSE Consortium has been funded by the Canadian Institutes for Health Research (CIHR) to develop guidance for the engagement of interest-holders in evidence synthesis and health guideline development. Additionally, members of the MuSE Consortium contribute to the development of new definitions, frameworks and guidance for health research. The current publications and overarching research program of the MuSE Consortium are summarized in Figure 1. This figure has been provided to demonstrate the ongoing and complementary research being conducted to inform the upcoming MuSE guidance. Articles included in this thesis are highlighted.

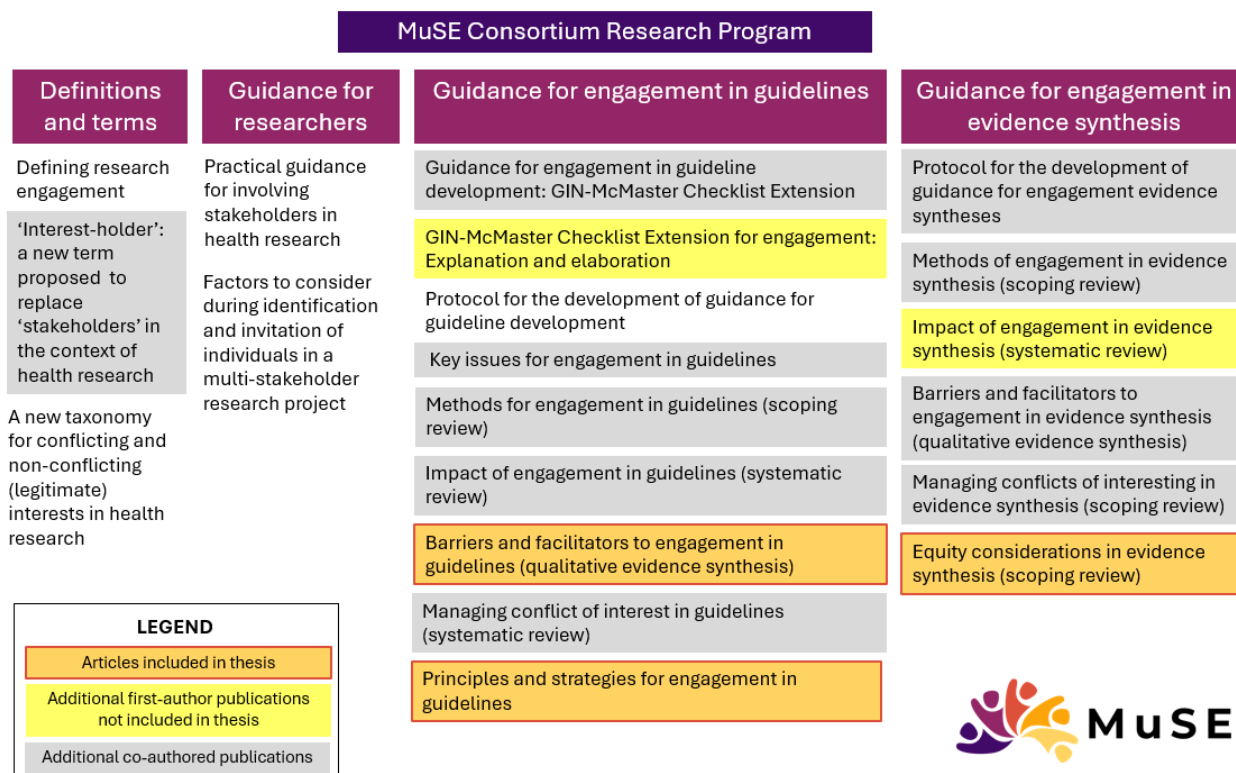


**Figure 1. Research program of the MuSE Consortium**

## 2.2 Interest-holders in health research

“Interest-holders” are groups of people whose interests should be considered in the context of health research and draw their legitimacy from the fact that these people are responsible for or affected by health- and healthcare-related decisions that can be informed by research evidence (Akl et al., in progress; Concannon et al., 2012). These include patients and caregivers, payers of health research, purchasers of health services, peer reviewed journal editors, policymakers, principal investigators, product makers, program managers, providers of care, and members of the public, referred to as the 11Ps (Petkovic et al., 2023; Tugwell et al., 2023) (See Table 1).

**Table 1: Interest-holders to engage in health research (Petkovic 2023; Tugwell 2023)**

| Broad category | MuSE 11P's                                                   | Description                                                                                                                            |
|----------------|--------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------|
| Researchers    | Principal investigators and all members of the research team | Individuals, organizations, and associations that conduct or advocate for health research, including the conduct of evidence syntheses |

|                         |                                                                |                                                                                                                                                                                                              |
|-------------------------|----------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Patients and the public | Patients, patient caregivers, patient advocates/ organizations | Those with lived experience with the condition of interest or who care for or advocate on behalf of those with lived experience                                                                              |
|                         | Public                                                         | Individuals in the general population of a defined geographic area excluding patients, caregivers, and health professionals living or working with the condition of interest                                 |
| Other professionals     | Payers of health research                                      | Individuals and organizations that fund research projects, such as government funders, industry funders, foundations                                                                                         |
|                         | Payers of health services                                      | Individuals, organizations and entities that pay for health services                                                                                                                                         |
|                         | Peer review editor                                             | Those who set journal policy on research publications (including evidence syntheses) and manage the peer review process and editing                                                                          |
|                         | Policymakers                                                   | Individuals, organizations and entities that craft public or private policy (on health) at any level of government                                                                                           |
|                         | Producers and commissioners                                    | Institutions and organizations that commission, develop, or implement evidence syntheses                                                                                                                     |
|                         | Product makers                                                 | Individuals working for companies that manufacture pharmaceuticals, medical devices, medical procedures, health technologies, or for profit educational and behavioural packages                             |
|                         | Program managers                                               | Managers/directors who plan, lead, oversee, or deliver any program that provides public health, community services, or clinical care (e.g. budgeting, hiring, staffing, organizing, coordinating, reporting) |
|                         | Providers                                                      | Persons and their professional associations who provide health care in a professional capacity or allowed by regulatory bodies to provide a health care service                                              |

Other common terms used to describe “interest-holders” include stakeholders, knowledge users, decision-makers, contributors, collaborators, partners, beneficiaries, interested parties, and others. While many of these terms are used across disciplines, they can be problematic when trying to describe the range of people or groups who could be considered for engagement in all health research. In part, because some terms (such as contributors, collaborators and partners) assume a pre-established relationship of engagement, but do not capture individuals with whom engagement is being considered but has not yet happened. Other terms, such as knowledge users or decision-makers, fail to recognize the people or groups who may produce knowledge or be affected by health decision-making. Finally, the term "stakeholder" has come under scrutiny for several reasons, leading to calls for its reconsideration in many contexts. One primary concern is its colonial roots in which a stakeholder was the person who drove a stake into the land to



demarcate the land they were stealing from Indigenous People (Reed et al., 2024; Reed & Rudman, 2024). This origin can be particularly insensitive when working with Indigenous communities or other groups that have been marginalized and dispossessed. By continuing to use "stakeholder," we risk perpetuating a mindset of ownership and control rather than fostering a spirit of partnership and shared purpose. Using an alternative term is more inclusive and accurately reflects the collaborative and mutual engagement necessary for population health research. The term "interest-holder" has been selected by the MuSE Consortium as a suitable replacement (Akl et al., under review), and has already been implemented by some groups (e.g., Sathe et al. 2024; Cochrane's People, Health Systems and Public Health Thematic Group, 2024).

## 2.3 Engagement

Engagement in health research refers to an approach to ensure the contribution of interest-holders towards the development, completion or dissemination of health research (Frank et al., 2020; Pollock et al., 2018). There are terms that have been used to describe engagement with specific interest-holders, including "public involvement", "consumer engagement", and "Patient and Public Involvement (PPI)". Other terms can describe engaging in partnership across interest-holder groups, such as "co-production", "co-creation", and "involvement". Table 2 lists some of the key terms and definitions to clarify their use.

**Table 2: Key terms and definitions relating to "engagement" (Todhunter-Brown et al., 2024; Magwood et al., 2024)**

| Term        | Definition                                                                                                                                                                                                                           | Notes on use                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                    |
|-------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Engagement  | A bi-directional relationship, or collaboration, between interest holders and a research team that results in informed decision-making about the selection, conduct, and use of research (Concannon et al., 2012; Vat et al., 2021). | <p>This is the preferred and most commonly used term in some parts of the world (including Canada and USA). It is therefore the term primarily used in this thesis</p> <p>It is important to note that the definition used here is the same as the definition of the term involvement used in other parts of the world (e.g. UK). In the UK, and some other parts of the world, engagement is considered something distinct from involvement, generally comprising interactions which may fall short of the definition we provide here. However, within this thesis, we adopt the Canadian/US usage, and we do not make any distinction between engagement and involvement.</p> |
| Involvement | Research being carried out 'with' or 'by' interest-holders (rather than research carried out 'about' or 'for' interest holders) (NIHR, 2021).                                                                                        | To accommodate international variations in terminology, we use the same definitions for the terms engagement and involvement.                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                   |

|                                                                                |                                                                                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                                                                                                                                                                                                                                                                                                    |
|--------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Co-production, and other terms starting with Co- (e.g. co-creation, co-design) | Internationally, there is no single, accepted definition of the term co-production, and it has been highlighted that anyone using this term should provide a definition (Masterton 2022). Co-production (and other co- terms) generally describe a process of engagement which adheres to a set of core values and principles. “Co-” terms often refer to people working together in ‘a partnership’ (Hoddinott 2018). | We consider these “co-”terms specific approaches which are related to, but which may go beyond, engagement. Sharing power and responsibility are key principles of these “co-” terms (Hickey 2018), potentially creating fundamental differences from some approaches to engagement. As definitions vary, we will not use these terms, instead describing the levels of engagement that interest-holders may have. |
| Partnership                                                                    | Like co-production, partnership describes a specific level of engagement in which particular values and principles are adhered to. It has been defined as “a relationship that involves mutual respect and an equal voice” (Hoddinott 2018).                                                                                                                                                                           | As above, we consider partnership a specific form of engagement. We will not use this term, but rather we will describe the levels of engagement that interest-holders may have.                                                                                                                                                                                                                                   |
| PPI (patient and public involvement)                                           | The term PPI refers to the involvement of patients and the public. Patients and the public are included within the interest-holder groups relevant to this thesis (Table 1)                                                                                                                                                                                                                                            | PPI is sometimes used as a noun, synonymous to involvement, sometimes referring to involvement of people and groups who are broader than “patients and public”. To ensure clarity we will avoid this abbreviation and describe the interest-holder groups who are engaged using the terminology in Table 1.                                                                                                        |
| Participatory research                                                         | An approach to research in which the goal is “to maximize the participation of those whose life or work is the subject of the research in all stages of the research process” (ICPHR, 2013).                                                                                                                                                                                                                           | Engagement of interest-holders in specific health research products (e.g., evidence syntheses and guidelines) are examples of participatory research.                                                                                                                                                                                                                                                              |

In recent years, enthusiasm for the science of participation in health service research has grown. For example, many funding organizations (e.g., Canadian Institutes for Health Research, National Institute for Health Research, Patient Centered Outcomes Research Institute) are increasingly promoting, or requiring, researchers to apply integrated knowledge translation (IKT) approaches to their research. IKT expects individuals or groups to be involved at various, or all, stages throughout the research process. For entities that produce evidence syntheses, for example, the engagement of interest-holders is already a formalized expectation (Cochrane, 2023; PCORI, 2024; Pollock et al., 2022; AHRQ, 2008).

Underpinning these commitments for engagement in research are epistemological, moral and consequentialist arguments (Boote et al., 2011). The epistemological argument states that interest-holders have direct experiential knowledge or expertise that can be of benefit to researchers, who may not have first-hand experience themselves of the health condition or

service that they are researching (Boote et al., 2011; Beresford, 2005). This represents a significant paradigmatic shift in what constitutes evidence for decision-making that has traditionally raised an epistemological tension between advocates for participatory paradigms and evidence-based paradigms (Palmer, 2020). Secondly, the moralistic argument states that interest-holders have a right to be involved in activities and decisions that may affect their health (Petkovic et al., 2020; Esmail et al., 2015) and that reducing the known power imbalances between researchers and groups such as patients is a moral duty of researchers, especially with oppressed and seldom-heard groups (Greenhalgh et al., 2019). Thirdly, the consequentialist argument states that engagement in research leads to more relevant research questions, improved study design, and more effective dissemination strategies, ultimately increasing the uptake and application of research outcomes, whilst also improving the transparency of the process and the accountability to the wider community (Boote et al., 2011; Domecq et al., 2014).

The goal of engagement is to ensure that research addresses the real-world needs and priorities of those it aims to serve, thus enhancing the relevance and impact of the findings. Engagement in health research fosters a sense of ownership and partnership among diverse interest-holders, promoting transparency and trust in the research process (Eales et al., 2017). It also helps to democratize knowledge production, ensuring that diverse perspectives and experiences inform research outcomes. By prioritizing engagement, health research can become more inclusive, equitable, and impactful, ultimately leading to improvements in healthcare quality and policy (Oliver et al., 2019).

### **2.3.1 Engagement for health equity**

Engagement of interest-holders can play a critical role in promoting health equity by ensuring that the voices of diverse groups, particularly those from marginalized communities, are heard and considered in decision-making processes. Health equity refers to the attainment of the highest level of health for all people, ensuring that everyone has a fair and just opportunity to be as healthy as possible (Braveman, 2014). In practice, achieving health equity requires addressing avoidable inequalities, injustices, and social determinants of health that create barriers for marginalized groups through the fair distribution of resources, opportunities, and outcomes

(Marmot, 2007; Whitehead, 1992). It requires deliberate efforts to ensure that resources and policies are distributed in ways that account for the varying needs of different populations (Whitehead, 1992). Engagement helps to address inequities in health by identifying structural and systemic barriers that contribute to health inequities, and fostering inclusive research, policies, and health systems that prioritize the needs of underserved populations. Equitable engagement focuses on the deliberate, inclusive, and meaningful engagement of diverse individuals or groups in decision-making processes or research activities, ensuring fair opportunities, access, and representation for all regardless of their backgrounds (Ul Haq et al., 2023). Equitable engagement aims to address disparities, promote inclusivity, and dismantle barriers that might prevent certain individuals or groups from actively participating or having their voices heard (NCCDH, 2021). By involving interest-holders from different communities in evidence synthesis research and health guideline development, health initiatives can be more reflective of the actual needs of diverse populations. This helps ensure that policies and interventions do not unintentionally exclude or harm groups.

Several frameworks for engagement exist which describe the different levels of engagement an interest-holder may have. The main difference between levels of engagement is the person's power, control, or decision-making authority. These include:

### **2.3.2 Arnstein's Ladder of Participation**

Arnstein's Ladder of Participation is a foundational theoretical framework for citizen involvement in decision-making, particularly in the context of social and public policy. The framework consists of eight rungs describing three general forms of citizen power: Nonparticipation (no power), Degrees of Tokenism (counterfeit power), and Degrees of Citizen Power (actual power) (Figure 2; Arnstein, 1969). Arnstein's Ladder emphasizes the need for genuine participation where citizens have actual power and influence over decisions that affect their lives. It highlights ways in which participatory processes can be superficial or manipulative and stresses the importance of striving for higher rungs where true power-sharing occurs.

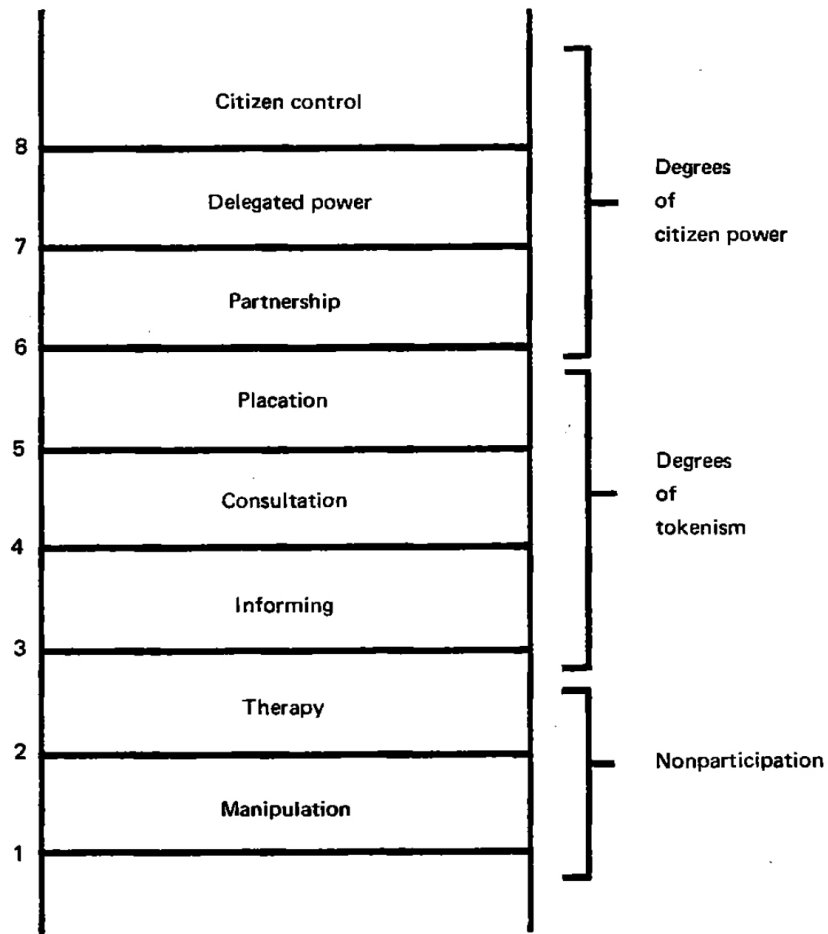


**Figure 2: 1969 illustration of Sherry Arnstein’s Ladder of Citizen Participation**

Although originally designed for social planning, the ladder has been applied in numerous fields including health and education. Arnstein emphasizes that genuine participation requires a transfer of power, encouraging policymakers to recognize and address power imbalances between “authorities” (e.g., policymakers) and citizens. While its focus on power imbalances is a strength of the model, it also frames participation in adversarial terms (citizens vs. authorities), which overlooks collaborative and mutually beneficial forms of engagement that do not fit neatly into the ladder's categories. Additionally, the ladder assumes a linear progression of participation from manipulation to citizen control, which may not accurately reflect the complexities of real-world participatory processes. Participation is often more fluid and context-dependent than the model suggests. While Arnstein’s Ladder remains influential, it may benefit from updates to reflect the complexities of contemporary engagement efforts.

### 2.3.3 IAP2 Spectrum of Public Participation

Developed by the International Association for Public Participation (IAP2), this spectrum outlines levels of public participation with increasing impact on decision-making and their associated ‘promise to the public’ (Figure 3; IAP2, 2018). Unlike Arnstein's Ladder, which focuses on power dynamics, the IAP2 Spectrum emphasizes the goals of participation at each level, making it easier for organizations to choose the most appropriate form of engagement based on their goals.

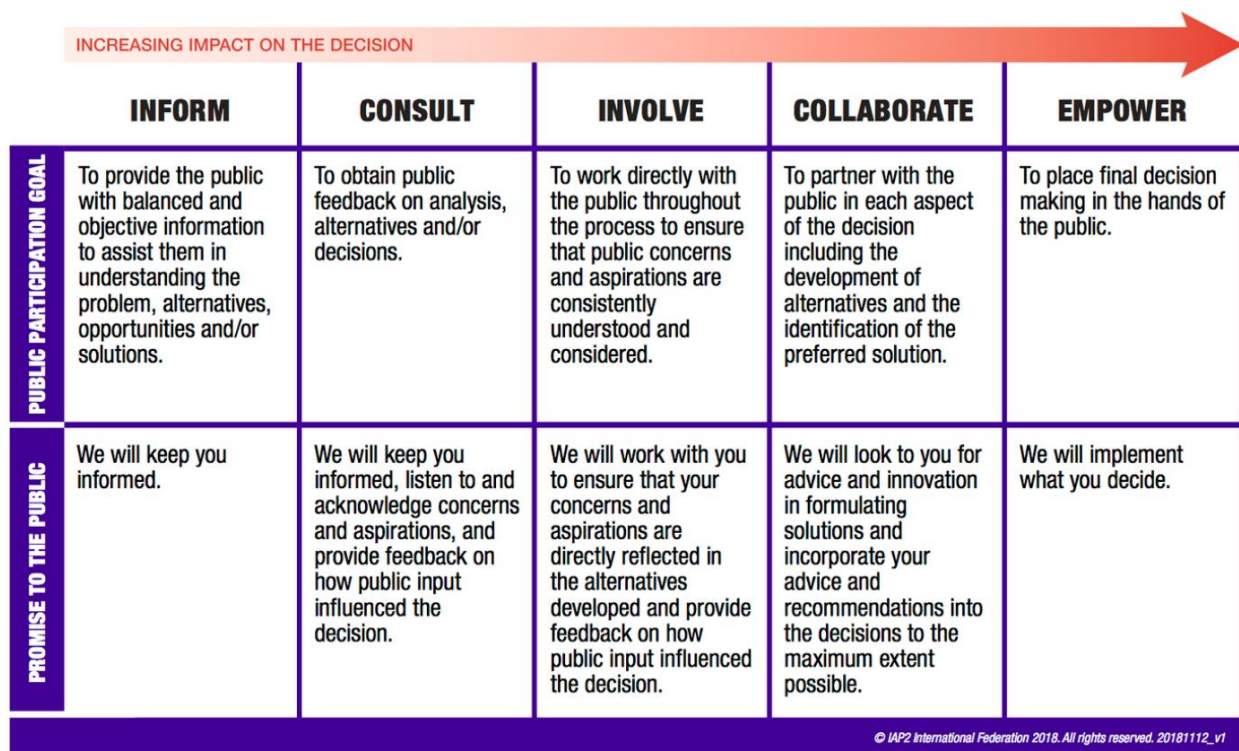


**Figure 3: IAP2’s Spectrum of Public Participation**

A key feature of the IAP2 Spectrum of Public Participation is its focus on the ‘promise to the public’. By setting clear expectations about how public input will be used, the framework promotes transparency and helps build trust among interest-holders. However, the spectrum does not explicitly address power imbalances between authorities and the public, which are often central to participatory processes. Unlike Arnstein’s Ladder, which frames participation as a struggle for power between authorities and citizens, the IAP2 Spectrum encourages collaboration and mutual decision-making. It recognizes that different levels of participation are appropriate depending on the context. Additionally, the IAP2 Spectrum does not explicitly address how to

engage marginalized or underrepresented groups. It assumes that public participation will engage all interest-holders, but without specific mechanisms to address equity, it risks excluding voices that are hardest to reach. For those seeking to address equity and power imbalances, it may need to be supplemented with additional tools or frameworks that explicitly address these issues.

### **2.3.4 4C's Model**

Previous engagement work identified 4 levels of engagement, termed the 4C's Model, adapted from other sources (Petkovic et al., 2023). The four levels of the model are: (1) Communication: Interest-holders receive information but have no role in contributing, (2) Consultation: Interest-holders provide their views, thoughts, feedback, opinions or experiences but without a commitment from the project team to act on them, (3) Collaboration: Interest-holders are engaged to influence the production of the project (e.g. commenting, advising, ranking, voting, prioritizing, and reaching consensus) without direct control over decisions, and (4) Coproduction: Interest-holders are equal members of the project team and have a key role in decision-making in the research process. By including levels such as collaboration and co-production, the 4C's Model encourages more inclusive decision-making processes where interest-holders have meaningful roles and influence. This can help build trust and ensure that decisions are reflective of interest-holder priorities. The higher levels of the model (collaboration and co-production) emphasize power sharing between interest-holders, which is essential for fostering more equitable outcomes. However, there is ambiguity between the transitions from one level to the next. For instance, distinguishing between consultation to collaboration may not be straightforward. Additionally, the highest level of engagement (co-production) can be difficult to achieve in practice. It requires significant resources, commitment, and often a shift in organizational culture to truly share decision-making authority with interest-holders. Meaningful collaboration and co-production involve a long-term commitment and might not be feasible for all organizations, especially those with limited resources. In some situations, co-production may not be feasible or desirable, and lower levels of engagement, such as consultation or collaboration, may still be meaningful and effective.

### 2.3.5 The ACTIVE Framework

The ACTIVE (Authors and Consumers Together Impacting on eVidencE) framework was developed to assist authors with reporting how and when people (referred to as “stakeholders”) were engaged in a systematic review (Pollock et al., 2019). It considers broad groups of interest-holders, namely patients, carers and family members, or any other ‘professional’ interest-holder. The framework includes the following constructs: (i) Who was involved?, (ii) How were stakeholders recruited?, (iii) What was the mode of involvement (approach and methods)?, (iv) At what stage in the review process did involvement occur?, and (v) What was the level of involvement (level of control or influence over the review process)? The levels of engagement are described as follows:

- Leading: Interest-holders initiate the review, and/or are the lead responsible for carrying out and completing the review.
- Controlling: Working in partnership with researchers, interest-holders have varying degrees of control or influence over the review process. They make decisions or control one or more aspects of the review process, in collaboration with or under the guidance of the review authors.
- Influencing: Interest-holders are stating, commenting, advising, ranking, voting, prioritizing, or reaching consensus. They provide data or information which should directly influence the review process, but without direct control over decisions or aspects of the review process.
- Contributing: Interest-holders provide views, thoughts, feedback, opinions or experiences. They provide data or information which may indirectly influence the review process. People may be participants in a research study (e.g. focus groups or interviews).
- Receiving: Interest-holders receive information about the systematic review, or results of the review.

The ACTIVE framework encourages interest-holders engagement to influence not only the research outcomes but also the way evidence is synthesized and communicated. However, without adequate support, resources, and training, patients and carers may struggle to contribute effectively, particularly in technical or complex areas such as systematic reviews. Researchers



may also face difficulties in integrating interest-holders input without overburdening the process or diluting scientific rigor. Power imbalances between researchers and patients/carers may persist. Researchers may unconsciously dominate decision-making processes due to their expertise in subject matter or technical areas. Bridging the gap between “laypeople” and experts requires significant effort to ensure that patients/carers feel empowered. While the ACTIVE framework emphasizes collaboration and impact, it lacks explicit mechanisms for measuring the effectiveness of engagement activities. Without clear metrics, it may be difficult to assess whether interest-holder engagement truly influences research outcomes or whether the framework’s goals are being met.

### **2.3.6. Levels of engagement of the MuSE Consortium**

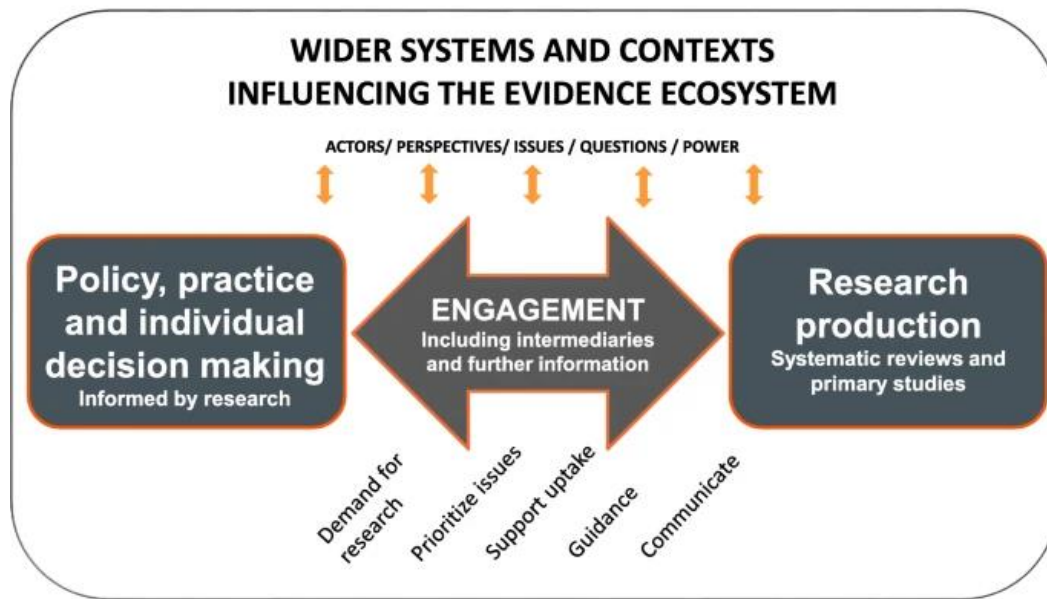
Within health research processes such as evidence synthesis and health guideline development, the level of engagement is expected to be flexible and may change from one activity to the next depending on the project needs and resources. The MuSE Consortium previously assessed papers reporting on interest-holder engagement in guidelines and identified challenges in operationalizing the 4C’s model and ACTIVE framework levels of engagement (Petkovic et al., 2023; Tugwell et al., 2023). Based on the detail provided in the studies assessed, the distinction between levels was unclear. Similarly, the details defining ‘collaboration’ from ‘coproduction’ were missing in published reports. Therefore, the MuSE Consortium opted to simplify this by categorizing engagement into two levels based on exploratory discussions with members of the MuSE Consortium and individuals representing interest-holder groups. The first level is ‘advice/feedback’, where interest-holders provide their opinions, perspectives, or experiences, which are considered by the project team. The second level of engagement is ‘decision-making’ whereby interest-holders actively contribute as an ‘equal voice’ to making decisions about research activities. While there are nuances within each of the two levels of engagement, the use of two clearly distinct levels is more feasible for the assessment of engagement in evidence synthesis and guideline development (Petkovic et al., 2023). Therefore, these two levels are referred to throughout this thesis.

## 2.4 Evidence ecosystems

“Evidence ecosystems” describes the relationship between interest-holder’s use of evidence to inform health decision-making (e.g., using health guidelines) and the production of evidence to inform these decisions in the form of evidence syntheses (Gough et al., 2019). Traditionally, ‘evidence producers’ such as principal investigators and members of their research teams were seen to operate independently from ‘evidence users’ who use research to inform health decision-making (e.g., policymakers, providers of care, patients). From a systems perspective (Best et al., 2010), evidence ecosystems illustrate that as users of evidence become more engaged with the evidence they use, they increasingly impact the production of that evidence (Gough et al., 2019). The basic components of an evidence ecosystem are depicted in Figure 4 and include the following (Gough et al., 2019):

- The use of research for informing decisions;
- Engagement of users and producers of research, providing access to and supporting interpretations of research and its uptake;
- Production of evidence syntheses (e.g., systematic reviews) and primary studies
- The broader socio-political context within which the evidence ecosystems of research use and research production exist.

# EVIDENCE ECOSYSTEMS



Adapted from (Gough et al 2019)

**Figure 4. Evidence ecosystems** (Gough et al., 2020).

Many tools may influence how evidence-informed decisions regarding health policy, clinical practice or personal health are made. Health guidelines, for example, are systematically developed statements to assist decision-makers which are developed through rigorous evidence synthesis, assessments of the benefits and harms of health interventions, and considerations of societal values and preferences (IOM, 2011). Evidence synthesis, therefore, plays a critical role in evidence ecosystems, balancing the needs of decision-makers with the availability of research and research production communities (Gough et al., 2020). For example, there are now over 10,000 published Cochrane systematic reviews and it is estimated that 75% of World Health Organization (WHO) guidelines cite Cochrane reviews (Cochrane, 2020). WHO has also called for health systems that are more dynamic and multisectoral, requiring “policies grounded in tangible evidence for ‘what works’ and how. This is a fundamental contribution of the research community - but it means providing research evidence that is synthesized, accessible and contextualized, to enhance its applicability in different health systems” (WHO, 2018).

This thesis examines engagement within this evidence ecosystem by studying the engagement in evidence syntheses and the development of health guidelines of 11 different groups of interest-holders (11Ps). For the purpose of this thesis, we refer to the following definitions:

**“Evidence synthesis”** refers to a research methodology using a systematic and rigorous approach of gathering, assessing, interpreting, and integrating available research evidence from multiple studies or sources to generate comprehensive and reliable conclusions. Many different types of evidence syntheses exist and are characterized by their research questions and analytic approaches. These include scoping reviews, rapid reviews, and quantitative or qualitative systematic reviews, among others (Magwood et al., 2024). In addition to the mainstream Cochrane-style quantitative systematic reviews and meta-analyses, evidence syntheses now include a range of alternative methods in which researchers make interpretative and value-laden judgments throughout the research process (Abrams et al., 2021).

**“Guidelines”** are systematically developed evidence-based actionable statements which assist interest-holders to make informed decisions about appropriate health interventions (WHO, 2003). The steps to consider for guideline development are outlined by the GIN-McMaster Guideline Development Checklist (GDC) (Schunemann et al., 2014), a comprehensive checklist of items that guideline developers could consider for all stages of the guideline enterprise, from planning the initiative, synthesizing the evidence, and formulating recommendations to implementation, evaluation and updating. The checklist comprises 18 overarching topics, several of which overlap with the evidence synthesis process.

## **2.5 Knowledge gaps in engaging interest-holders in evidence synthesis and health guidelines**

Two recent frameworks are available to guide engagement in evidence synthesis: The Haddaway Framework for engagement during systematic reviews and maps in environmental management (Haddaway et al., 2017) and the ACTIVE Framework for involvement in systematic reviews (Pollock et al., 2019). Both frameworks primarily focus on systematic reviews rather than broader forms of evidence synthesis and have some limitations in regard to their applicability to interest-holders in the field of health. The Haddaway Framework presents some preliminary methodological options for engaging interest-holders, but it is only partially relevant to the field of health and health care. In environmental evidence syntheses, the types of interest-holders can vary significantly depending on the enormously varied research questions that environmental

studies assess (Haddaway et al., 2017) . In health care, the types of interest-holders remain much more constant regardless of the research question, as evidenced by multiple papers reporting nearly identical classifications of interest-holders (Concannon et al., 2012; Rader et al., 2014; Tugwell et al., 2006). The ACTIVE Framework was designed as a descriptive framework to describe interest-holder involvement in completed published systematic reviews for a subset of interest-holders relevant to health, namely patients, carers and families, and “others” (including health professionals, health decision makers and funders) (Pollock et al., 2019). Additional interest-holders defined by the MuSE 11P’s (e.g., peer-review journal editors, product makers, payers of health services) may have very different issues for evidence synthesis and their engagement may have different implications. There is a need to update and operationalize these frameworks to provide detailed instructions on it when planning and conducting a broad range of evidence syntheses such as scoping reviews, rapid reviews and qualitative evidence synthesis.

Additionally, neither framework explicitly aimed to be equity-oriented or be developed through a health equity lens. As funding agencies are increasingly promoting the use of engaged research approaches, additional guidance may be needed to support researchers and interest-holders in establishing and conducting research equitably (Hoekstra et al., 2020). The MuSE Consortium aims to develop equity-oriented guidance on methods for conducting, evaluating, and reporting engagement in evidence syntheses (Tugwell et al., 2023).

Complementary guidance is also being developed by the MuSE Consortium for the meaningful and equitable engagement of multiple interest-holders in health guideline development (Petkovic et al., 2020). Presently, planning tools such as the Guidelines International Network (GIN)-McMaster Guideline Development Checklist (GDC) exist but include only a few items about interest-holders engagement, in part because it is a more recent phenomenon. The MuSE Consortium guidance aims to incorporate interest-holder engagement as a cross-cutting consideration for each topic of the checklist. While some guidance for the engagement of patients and the public in guideline activities exist, such as the GIN-Public Toolkit (<https://g-i-n.net/toolkit>), Armstrong and colleagues’ (2017) framework for enhancing clinical practice guidelines through continuous patient engagement, and the EVOLVE framework for meaningful patient involvement in clinical practice guideline development and implementation (Bjorkqvist et

al., 2021), the existing guidance is insufficient. Firstly, all three resources focus on the engagement of patients and members of the public, but do not consider other distinct groups of interest-holders identified by the MuSE Consortium's 11Ps. There are several key issues for other groups of interest holders, such as conflict of interest concerns, which need to be managed throughout guideline development (Petkovic et al., 2023). Secondly, the recommendations for engagement are inconsistent between these three frameworks. Particularly, the EVOLVE framework does not consider patient engagement in the systematic review to be a priority for guideline developers (Bjorkqvist et al., 2021). However, these evidence reviews form the basis of decision-making in guideline development, and the exclusion of patients from such processes may overlook patient priorities and contribute to perpetuating health inequities. Finally, none of the available guidance considers engagement in the organization, planning and budgeting of guideline development initiatives. Engagement of interest-holders in these early stages of guideline development can help ensure that the goals of engagement have been elucidated and that proper resources are available to meet those goals. Overall, there is a need for comprehensive guidance which considers the engagement of diverse and distinct groups of interest-holders in all stages of guideline development. Understanding the barriers and facilitators to engagement is essential for the development of this comprehensive guidance. Currently, most of what is known about barriers and facilitators to engagement is specific to patients and members of the public (Legare et al., 2011).

To support future engaged research approaches and based on a review of partnership literature by Hoekstra and colleagues (2020), additional research should focus on the development of evidence-based tools and resources for engaged research related to principles and strategies for engagement. Principles are rules or statements that describe well-established, widely accepted, overarching ideals for engagement which establish a desirable course of action (Pinho-Gomes et al., 2022). Strategies underpinning engagement should enact those principles in the form of guidance or frameworks. To complement the guidance from the MuSE Consortium and promote its operationalization, it is critical that principles and strategies for engaging interest-holders in guideline development are identified and acceptable to interest-holders themselves. Conducting interviews with interest-holders can provide deep insights into their experiences, beliefs, and attitudes about guideline development. This qualitative data can uncover nuanced perspectives

and contextual factors influencing their engagement. Therefore, primary data collected from interest-holders is needed to complement evidence available from the published literature to inform the MuSE guidance for engagement in guideline development.

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## CHAPTER 3: ARTICLE 1. Considerations for the equitable engagement of patients and other interest-holders in evidence synthesis: A scoping review

### Chapter Overview

This Chapter presents a scoping review on equity considerations for the engagement of interest-holders in evidence synthesis research. The article has been prepared for publication in *Cochrane Evidence Synthesis and Methods* in an upcoming special collection.

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### 3.1 ABSTRACT

**Background:** Evidence syntheses such as systematic or scoping reviews are the most common product to inform decision making for health policy and practice. Meaningfully engaging patients and other interest-holders in the evidence synthesis process is gaining momentum, but there is a need for equity-related guidance for this type of collaborative research approach. This scoping review aims to identify and characterize the equity considerations for the engagement of eleven different groups of interest-holders in evidence synthesis.

**Methods:** We searched Medline, EMBASE, PsycINFO, CINAHL and SCOPUS from database inception up to 12 January 2024. We included articles that described the engagement of 11 interest-holder groups such as patients, providers, and policymakers in the evidence synthesis process. We identified equity considerations for their engagement across PROGRESS-Plus characteristics and summarized principles to support equitable engagement.

**Results:** We included 93 studies (published in 97 reports) in this scoping review. The perspectives of patients, providers and members of the public were most commonly considered. Engagement in evidence synthesis is concentrated in high-income English-speaking countries. Issues related to access to technology can result in resource bias and exclude people from lower socio-economic positions. We identified considerations for engaging with youth and people living with disabilities, such as tailored training and support. Further, we identified four principles to support equitable engagement: Addressing power dynamics, accessibility, promoting diversity and inclusion, and ethical considerations.

**Conclusion:** There are opportunities to equitably and meaningfully engage all groups of interest-holders in evidence synthesis methods. Engaging diverse interest-holders may promote social justice and shape more equitable health systems. Future research should explore the equitable engagement of interest-holders from low-income settings, diverse language backgrounds, and different socio-economic positions.

**Keywords:** Patient and public involvement, stakeholder engagement, interest-holder, evidence synthesis, equity

### 3.2 INTRODUCTION

Evidence synthesis is the systematic and rigorous approach of gathering, assessing, interpreting, and integrating available research evidence from multiple studies or sources to generate comprehensive and reliable conclusions. Common evidence synthesis methods include systematic reviews, scoping reviews, realist reviews, rapid reviews, qualitative evidence synthesis, and evidence maps. These syntheses can provide a robust foundation for decision-making in healthcare, policy formulation, and other fields. Evidence synthesis, therefore, plays a critical role in evidence ecosystems, balancing the needs of decision-makers and end-users with the availability of primary research and research production communities (Gough et al., 2020; Ravaud et al., 2020). This raises broad concerns about societal engagement with research, such as power and agency, reflected by who sets research agendas, who is involved in evidence synthesis, and whether the scope, data, analysis, findings, and recommendations sufficiently consider equity issues (Gough et al., 2020). Failing to include certain groups in society from evidence synthesis research can have repercussions on resource allocation and perpetuate cycles of inequality (Dewidar et al., 2022; Gough et al., 2020; Kelly et al., 2015; Welch et al., 2013).

Integrating patients, members of the public, and other interest-holders (particularly those who are traditionally excluded from research) into evidence synthesis is becoming the “new normal” (Beames et al., 2021). “Interest-holders” are groups of people whose interests should be considered in the context of health research. The interests arise and draw their legitimacy from the fact that these people are responsible for or affected by health- and healthcare-related



decisions that can be informed by research evidence (Akl et al., in progress). The MuSE Consortium has identified 11 categories of interest-holders who could potentially be engaged in evidence synthesis research: Patients, payers of health research, purchasers of health services, peer reviewed journal editors, policymakers, principal investigators, producers and commissioners of reviews, product makers, program managers, providers of care, and members of the public (Tugwell et al. 2023). These groups have previously been termed “stakeholders”, however we recognized a number of negative historical uses of the word "stakeholder" such as its mercenary roots as the person who physically held the stakes in a bet (Sharfstein 2016) or its colonial roots in which a stakeholder was the person who drove a stake into the land to demarcate the land they were stealing from Indigenous territories (Reed & Rudman, 2022). This change in terminology represents an intentional shift towards equitable research engagement practices.

The engagement of relevant interest-holders can produce a more contextually valid synthesis relevant in situations characterized by an equity context, and, therefore, may enhance considerations of equity in health decision-making and implementation (Harris et al., 2016; Akl et al., 2017). Health equity is the absence of systematic disparities in health between social or demographic characteristics (Braveman & Gruskin 2003), such as those outlined by the PROGRESS mnemonic: place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, and social capital (O’Neill et al., 2014; Evans & Brown 2003). Oliver *et al.* (2008) expanded this framework to ‘PROGRESS-Plus’ to include other factors related to inequity, such as sexual orientation, age, and disability. Achieving health equity involves eliminating disparities to ensure that all individuals can attain their highest level of health through the fair distribution of resources, opportunities, and outcomes (Marmot, 2007). To truly address social inequities, engagement in research processes such as evidence synthesis should go beyond the mere inclusion of certain societal groups in research processes, which risks being tokenistic (Arnstein, 1969). Equitable engagement focuses on the deliberate, inclusive, and meaningful engagement of diverse individuals or groups in decision-making processes or activities, ensuring fair opportunities, access, and representation for all participants regardless of their backgrounds or circumstances (Ul Haq et al., 2023). Equitable engagement aims to address disparities, promote inclusivity, and dismantle barriers

that might prevent certain individuals or groups from actively participating or having their voices heard (NCCDH, 2021).

To date, there is a lack of available guidance to assist researchers, decision-makers, and other interest-holders in engaging in the evidence synthesis process equitably. The MuSE Consortium aims to address this gap by developing equity-oriented guidance on methods for conducting, evaluating, and reporting engagement in evidence syntheses (Tugwell et al., 2023). As part of the Consortium's work, a series of five evidence syntheses are underway which examine: 1) Engagement methods, 2) Barriers and facilitators to engagement, 3) Impact of engagement, 4) Conflict of interest management, and 5) Equity considerations. This scoping review of equity considerations represents the fifth review in this series, and a protocol outlining the series has been published elsewhere (Tugwell et al., 2023).

### **3.3 RESEARCH OBJECTIVES**

The objectives of this scoping review are:

1. To critically examine the definitions of “interest-holder” and “engagement” in the evidence synthesis literature;
2. To identify existing guidance, frameworks, models and checklists for equity considerations for engagement during the conduct of evidence syntheses; and
3. To summarize and map equity considerations for engagement across equity characteristics (defined by PROGRESS-Plus; O'Neill et al., 2014).

### **3.4 METHODS**

We developed a protocol and conducted a scoping review according to published guidance (Arksey & O'Malley, 2005; Levac et al., 2010; Peters et al., 2022). We reported our review according to the PRISMA extension for scoping reviews (PRISMA-ScR; [Tricco et al., 2018]) [Supplementary File 1]. We reported our search strategy according to the PRISMA

Statement for Reporting Literature Searches in Systematic Reviews (PRISMA-S; [Rethlefsen et al., 2021]) [Supplementary File 2].

### 3.4.1 Eligibility criteria

We included articles which met the following eligibility criteria, defined by the PCC framework (Population, Concept and Context) (Peters et al., 2022).

**Population:** We included articles that described interest-holders organized by the following 11Ps: patients and caregivers, the public, providers of care, policymakers, program managers, payers of health research, payers of health services, peer-reviewed journal editors, and product makers. Table 1 describes each population (Petkovic et al., 2023; Tugwell et al., 2023).

**Table 1. Groups to consider engaging in evidence synthesis (Tugwell et al., 2023)**

| Groups                                                        | Description                                                                                                                                                                     |
|---------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Patients, patient caregivers, patient advocates/organizations | Those with lived experience with the condition of interest or who care for or advocate on behalf of those with lived experience                                                 |
| Payers of health research                                     | Individuals and organizations that fund research projects, such as government funders, industry funders, or foundations                                                         |
| Payers / Purchasers of Health Services                        | Individuals, organizations, and entities that pay for health services                                                                                                           |
| Peer-Reviewed Journal Editors                                 | Those who set journal policy on guidelines and manage the peer review process and editing                                                                                       |
| Policymakers                                                  | Individuals, organizations, and entities that craft public or private policy (on health) at any level of government                                                             |
| Principal Investigators and all members of the research team  | Individuals, organizations, and associations that conduct or advocate health research                                                                                           |
| Producers and commissioners of reviews                        | Institutions and organizations that commission, develop, or implement evidence synthesis or guideline development procedures                                                    |
| Product makers                                                | Individuals working for companies that manufacture pharmaceuticals, medical devices, medical procedures, health technologies, or for-profit educational and behavioral packages |

|                  |                                                                                                                                                                                                              |
|------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Program managers | Managers/directors who plan, lead, oversee, or deliver any program that provides public health, community services, or clinical care (e.g. budgeting, hiring, staffing, organizing, coordinating, reporting) |
| Providers        | Persons and their professional associations who provide health care in a professional capacity and are allowed by regulatory bodies to provide a health care service                                         |
| Public           | Individuals in the general population of a defined geographic area, excluding patients, caregivers, and health professionals living or working with the condition of interest                                |

**Concept:** Articles must report on the engagement of at least one group, defined in Table 1 during the conduct of an evidence synthesis. “Engagement” can include providing advice or feedback (where opinions, perspectives, experiences, or values are sought and considered by the review team) or participating in decision-making (whereby interest-holders actively contribute as an “equal voice” to making decisions about review activities) (Petkovic et al., 2023). Articles may refer to any evidence synthesis product, including systematic reviews, scoping reviews, realist reviews, rapid reviews, qualitative evidence synthesis, and evidence maps.

**Context:** We included articles that described 1) recommendations or guidance for engaging with interest-holders during an evidence synthesis, 2) barriers or facilitators to engaging with interest-holders, 3) evaluations of the impact of engaging with interest-holders, and 4) equity considerations for engagement with interest-holders. These descriptions could be based on the author's personal experiences conducting engaged research, or collected using empirical methods.

**Study designs:** We included articles with the following broad range of study designs: Existing systematic or scoping reviews describing engagement in evidence synthesis, guidance or methodology papers describing recommended processes for engagement, impact evaluations of engagement in evidence synthesis, qualitative studies describing experiences of interest-holders, descriptive studies about evidence synthesis engagement activities, and commentaries by authors outlining their expertise and experiences. We excluded conference abstracts and posters. We also excluded individual evidence reviews on various health-related topics which may have engaged

interest-holders, as these are being analyzed in a separate review by the MuSE Consortium (Todhunter-Brown et al., 2024). We excluded articles published in languages other than English, French and Spanish.

### **3.4.2. Search methods**

We developed our search strategies in consultation with a health sciences librarian (L.H.). We searched the following databases, individually, from database inception up to 12 Jan 2024: Medline (Ovid), EMBASE (Ovid), PsycINFO (Ovid), CINAHL (Ebsco) and SCOPUS. We used a combination of keywords and subject headings to combine concepts of “engagement of interest-holders” (e.g., patients, end-users, co-production) and “evidence synthesis” (e.g., systematic review, meta-analysis). Complete search strategies for each database are available in Supplemental File 3. We also conducted a focused gray literature search by searching the websites of the following organizations: Cochrane, Patient-Centered Outcomes Research Institute (PCORI), Canada’s Strategy for Patient-Oriented Research (SPOR), Cochrane Consumers Network, JBI, INVOLVE, World Health Organization (WHO), National Institute for Health and Care Excellence (NICE), Stakeholder Engagement in Evidence Synthesis, and the Guidelines International Network (GIN). Members of the MuSE Consortium could also recommend literature for consideration.

### **3.4.3. Screening and selection**

We used Covidence software (Veritas Health Innovation) to deduplicate search results and facilitate a two-stage study selection process: (1) a title and abstract review, and (2) full-text review. Two review authors independently assessed all potential articles against a priori eligibility criteria. The lead author (O.M.) screened all studies at the full-text stage alongside a second reviewer. We resolved any disagreements through discussion, or we consulted a third review author.

#### **3.4.4. Data extraction and management**

We used a standardized extraction sheet. Pairs of reviewers extracted data in duplicate and independently using Covidence. The lead author (O.M.) extracted all studies alongside a second reviewer. They compared results and resolved disagreements by discussion or with help from a third reviewer. We extracted the following information: Study objective, study design and methods, type of evidence synthesis, the definition of interest-holder and specific groups identified, the definition of engagement, funding, and conflicts of interest statements. Furthermore, we characterized whether the articles reported recommendations for engagement, an impact assessment or evaluation, barriers or facilitators to engagement, or equity considerations. For all studies reporting equity considerations, we categorized the findings according to PROGRESS-Plus (O'Neill et al., 2014): Place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socio-economic status, and social capital; for the 'Plus' we extracted age and disability. We also extracted any additional statements related to principles that promote equitable engagement.

#### **3.4.5. Synthesis of results**

We tabulated the characteristics of all included studies and visualized the counts of interest-holder groups represented in the included studies using bar charts using Microsoft Excel. To identify the most common terms used to describe "interest-holders" and "engagement", we conducted frequency analyses using NVivo (QSR International) and presented word clouds. Two review authors (O.M., V. H.) summarized categorized equity-relevant statements across PROGRESS-Plus characteristics and thematically identified key principles for equitable engagement through a content analysis, counting the occurrence of concepts or keywords to infer meaning (vs. thematic analysis which assigns meaning by extracting high-level ideas) (Forman & Damschroder, 2007). The content analysis consisted of iteratively developing a coding scheme based on a subset of studies, and then applying the coding scheme to all articles which reported equity considerations. Two authors (O.M. and V.H.) read all included articles and came to agreement about which articles reported equitable engagement principles. One author (V.H.)

developed the initial coding categories (n=7). A second author (O.M.) re-read all articles and suggested the collapse of similar categories (e.g., Power-sharing and Empowerment), the removal of categories which were inadequately supported (e.g., Autonomy and Self-Determination) and the addition of a new category (Ethical considerations). All concepts were reviewed by the authorship team and the wording was revised as needed.

### 3.4. RESULTS

#### 3.4.1 Results of the search

Our systematic search identified 113,786 citations. After the removal of duplicates, we screened 54,721 citations by title and abstract. We retained 788 for full text review. Of these, 81 studies (published in 85 reports) met full eligibility criteria. Additionally, we screened 127 documents identified through our gray literature search, and included 11. One additional article was identified by a member of the MuSE Consortium for inclusion. In total, we included 93 studies (in 97 reports) in this scoping review.

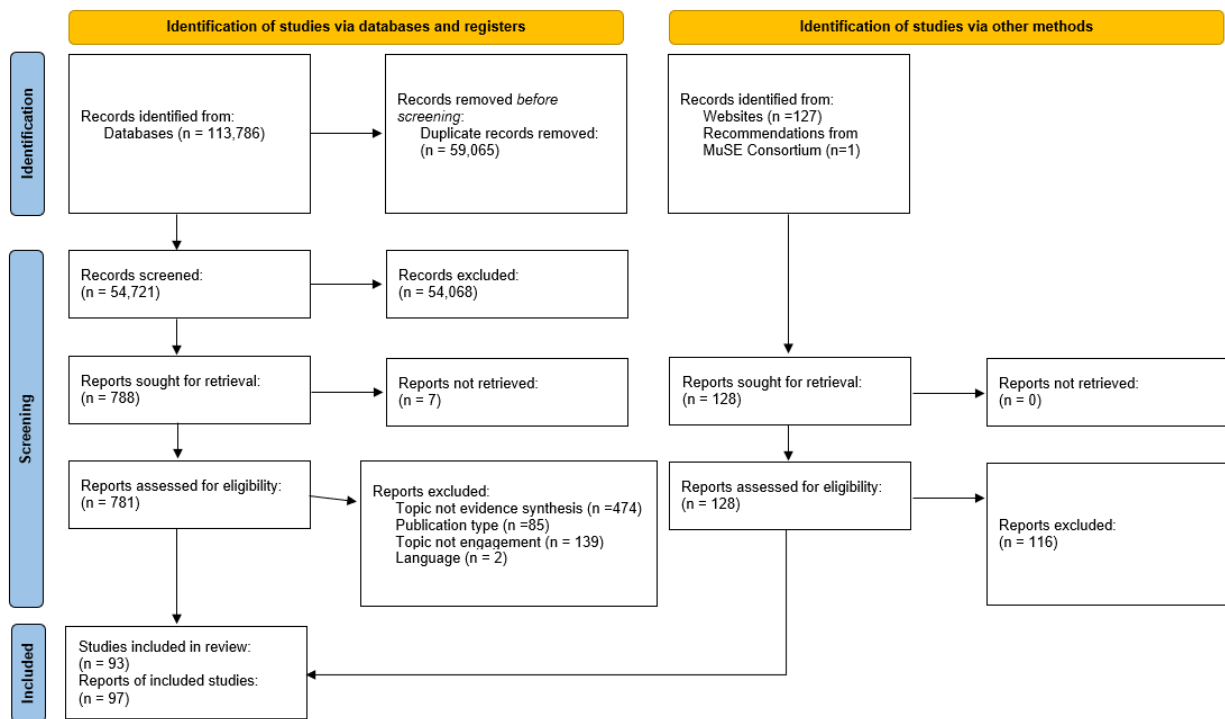


**Figure 1.** PRISMA flow diagram

### 3.4.2 Characteristics of included studies

We summarized the characteristics of all included studies in a table (Supplemental File 4). Among the 93 included studies, the majority reported on the engagement of interest-holders in systematic reviews (n=65). However, several other evidence synthesis review types were reported on, including rapid reviews (n=7), evidence maps (n=6), scoping reviews (n=4), realist reviews (n=3), evidence reviews for health technology assessments (n=4), and qualitative evidence synthesis (n=1). Eleven articles reported on approaches applicable to multiple review types or any type of evidence synthesis method. In the included articles, interest-holders from all 11P groups were represented, the most frequent being patients (n=72), providers (n=58), policymakers (n=48) and members of the public (n=46) (see Figure 2). We included the following study designs: Descriptive studies (case studies or cross-sectional studies) about evidence synthesis engagement activities (n=35), existing reviews describing engagement in evidence synthesis (n=20), guidance or methodology papers describing recommended processes for engagement (n=14), empirical qualitative or mixed method studies on experiences of engaging interest-holders (n=9), impact evaluations of engagement in evidence synthesis (n=8), and commentaries by authors outlining their expertise and experiences (n=7). Research primarily originated from high-income countries such as the UK, USA, Canada, Australia or across Europe. None of the methodological or evaluative studies were conducted by evidence synthesis teams located in low and middle-income countries. One systematic review was conducted by a team based in Lebanon with funding from WHO (Fadlallah et al., 2020), and one commentary reported author experiences from the Africa Centre for Evidence at the University of Johannesburg, South Africa, funded by the UK's Natural Environment Research Council (Langer et al., 2017).



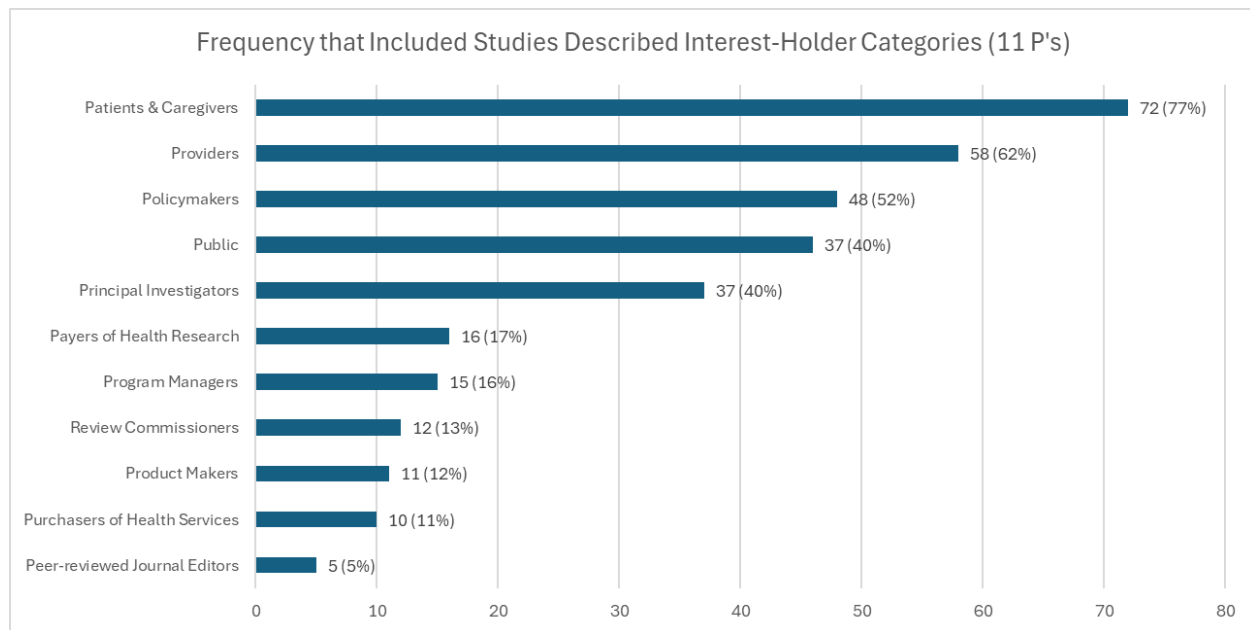


**Figure 2.** Frequency of interest-holder groups described among included studies. Percentages were calculated using the number of included studies (n=93) and rounded to the nearest integer.

### 3.4.3 Definitions of “interest-holder”

The term “stakeholder” remains the most frequently used term to describe interest-holders in evidence synthesis (Figure 3). This term was either used without a formal definition, or referred to those who are responsible for or affected by health-and healthcare-related decisions that can be informed by research evidence (Anderson et al., 2021; Buckley et al., 2014; Concannon et al., 2014; Cottrell 2014; Agyei-Manu 2023; Fadlallah et al., 2020; Springs et al., 2019; Todhunter-Brown et al., 2022). Articles often emphasized specific interest-holder groups, including patients, principal investigators and researchers, and the public. Notably, none of the included studies acknowledged the problematic historical connotations associated with the term stakeholder or the potential harms associated with its continued use. The use of other terms also appeared to be associated with the country of the authorship team. For example, the term ‘knowledge user’ was used primarily reported in Canadian (e.g., Tricco et al., 2018; Soobiah et al., 2019; Sanderson et al., 2022; Garritty et al., 2023) and Australian contexts (Pollock et al., 2022), whereas ‘patient and public involvement (PPI)’ was frequently used in literature

originating from the UK (e.g., Troya et al., 2019; Walker et al., 2021; Todhunter-Brown et al., 2022). The term “consumer” was also consistently used by the review-producing Cochrane Collaboration when referring to patients, carers and members of the public, They referred to ‘stakeholders’ when broadening to health care providers and policymakers.



**Figure 3.** Most frequent terms used to describe interest-holders

#### 3.4.4. Definitions of “engagement”

The most frequent term used to describe working with interest-holders in the conduct of evidence syntheses was “involvement”, often used in the phrase “patient and public involvement” (Figure 4). This was closely followed by the term “engagement”. Many articles used the terms “involvement”, “engagement” and “participation” interchangeably. They recognized that these terms refer to a range of activities spanning from communication between interest-holders to co-production of evidence syntheses. One document specified the differences between these terms, stating that involvement in research refers to research being carried out ‘with’ or ‘by’ members of the public rather than ‘to’, ‘about’ or ‘for’ them” (Cochrane Collaboration, 2022). This differs from engagement, which might include raising awareness of research, sharing knowledge, or engaging and creating a dialogue with the public (Cochrane Collaboration, 2022). Three articles also qualified approaches as “meaningful engagement”, described as the planned, supported and valued involvement of interest-holders (Boden et al., 2021), moving more towards cooperation and collaboration (Cooper et al., 2020). Meaningful

engagement is likely when the research process achieves high levels of trust, respect, accountability, legitimacy, competency, and fairness between interest-holders (Soobiah 2019). Engagement was also described as an “active partnership” (Tomlinson et al., 2021) and a bi-directional relationship that results in informed decision-making (Concannon et al., 2014; Land et al., 2017), leading to two-way information flow (Eales et al., 2017). Seven articles acknowledged the growing interest in “co” approaches to evidence synthesis, such as co-production, co-creation and co-design (Anderson et al., 2021; Heenan et al., 2023; Langer et al., 2017; Morley, 2022; Petticrew et al., 2023; Springs et al., 2019; Synnot et al., 2019). The underlying principles of these approaches were described as sharing of power and responsibility, including all perspectives and skills, respecting and valuing contributors’ knowledge, and cultivating mutually beneficial and supportive relationships (Synnot et al., 2019). Finally, several Canadian publications specified that their approach was “integrated knowledge translation (iKT)” (Harrison et al., 2012; Soobiah et al., 2019, Mogo et al., 2020, Sanderson et al., 2022). While multiple articles presented equity considerations, none of the included articles used the term “equitable engagement” when referring to their engagement processes.



**Figure 4.** Most frequent terms used to describe engagement

### 3.4.5. Considerations for equitable engagement

From 93 articles, 32 (34.4%) presented explicit considerations for equitable engagement. We have organized these considerations into two sections: equity considerations across PROGRESS-Plus characteristics (See Supplemental 5 and summary in Table 2), and principles that promote equitable engagement (Table 3).

### **Equity considerations across PROGRESS-Plus characteristics**

We identified a limited number of studies (n=17; 18.3%) that explicitly considered the engagement of interest-holders according to PROGRESS-Plus characteristics (See Supplemental 4). These articles reported on engagement considerations for individuals living in different places of residence (n=4), race/ethnicity/culture/language (n=8), occupation (n=3), gender or sex (n=2), socio-economic status (n=3), age (n=3), and living with disability (n=4) (See Table 2). In terms of geographical representation, there is a significant imbalance in interest-holder engagement. One study conducted by the Cochrane Collaboration reported predominantly engaging consumers (patients/public) from developed countries due to barriers to engagement for those in low- and middle-income countries (Morley et al., 2016). Authors of one review speculated that in these settings, the capacity for producing systematic reviews is limited and often misaligned with policy needs and priorities (Fadlallah 2020). Language and cultural differences pose challenges (Cochrane Collaboration, 2015; Morley et al., 2016), and patients in particular may provide stronger contributions if able to provide input in their local language (Bayliss et al., 2016; Arnstein et al., 2020). Two studies suggested that priority populations, such as gender or sexual minorities, may not be easily recruited to engage in relevant syntheses, leading to potential gaps in research perspectives (Troya et al., 2019; Boote et al., 2011).

Access to technology and other resources, such as the internet, is a concern, as interest-holders with lower access may face challenges responding to invitations or fully engaging in the evidence synthesis process (Haddaway et al., 2017). Representation of youth is highlighted as crucial, given their typical underrepresentation in the literature base, emphasizing the importance of empowering their voices (Cooper et al., 2020; Walker et al., 2021). Consent considerations, including caregiver consent for individuals under 16 years, and age-appropriate language, are also emphasized, highlighting the need for ethical considerations in engaging different age

groups (Cooper 2020 et al.,; Walker et al., 2021). Workshops and tailored training can enhance the involvement of interest-holders of diverse abilities (Brütt et al., 2017), while sensitivity to health conditions is crucial to avoid worsening mental or physical health during discussions (Walker et al., 2021). Providing interest-holders with multiple modes of interaction (for example, workshops, email, face-to-face interviews, etc.) can mitigate access and resource bias (Haddaway et al., 2017).

**Table 2: Summary of equity considerations across PROGRESS-Plus characteristics**

| Equity characteristics                                                 | Considerations for engagement                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
|------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Place of residence<br>( <i>n=4 studies</i> )                           | Four articles discussed differences in engagement between high-income and low- and middle-income countries (Morley et al., 2016; Dennett et al., 2022; O'Connor et al., 2023; Fadlallah et al., 2020). Consumer (patient/public) engagement in the Cochrane Collaboration is largely concentrated in developed countries, with barriers such as information scarcity and lack of facilities hindering engagement in developing nations (Morley et al., 2016). Efforts to address this disparity include hosting online workshops to attract diverse international participants (O'Connor et al., 2023), involving interest-holders in the review priority-setting process to align systematic review production with policy needs and priorities (Fadlallah et al., 2020), especially in resource-constrained settings, and retaining topics prioritized by individuals from low- and middle-income countries (Dennett et al., 2022).                                                                                          |
| Race, ethnicity, culture, language, ancestry<br>( <i>n=8 studies</i> ) | Five studies reported on language and cultural challenges in engaged evidence synthesis research (Arnstein et al., 2020; Munthe-Kaas et al., 2022; Bayliss et al., 2016; Morley et al., 2016; Cochrane Collaboration, 2015). Cochrane review groups in non-Western countries encounter barriers to consumer involvement, including language and cultural disparities (Cochrane Collaboration, 2015; Morley et al., 2016). Patient authors can contribute more effectively if they can provide input in their native language, especially when patients need support in understanding terminology and research processes (Arnstein et al., 2020; Bayliss et al., 2016). Tools such as the TRANSFER conversation guide could enhance engagement if available in the interest-holders' own languages (Munthe-Kaas et al., 2022). Three studies reported on race or ethnicity, indicating an overwhelming lack of representation from non-White interest-holders (Cornman & White, 2017; Synnot et al., 2019; Troya et al., 2019). |
| Occupation<br>( <i>n=3 studies</i> )                                   | All eleven groups of interest-holders were represented in the available literature to some degree (see Figure 2), many of which are related to occupations such as healthcare providers and policymakers. However, no studies reported on equity-related concerns regarding these occupations such that the individual would be put at a disadvantage for opportunities for health (such as discrimination bias, unstable employment, labor-intensive jobs, or holding temporary work permits). In the included studies, patient/caregivers were engaged in evidence synthesis research on the basis of a diagnosis or lived experience of a health condition, but their occupations were only reported in three studies (Coon et al., 2016; Hannigan et al., 2022; Synnot et al., 2019). The overlap between lived experience and occupational experience relevant to the evidence synthesis topic was seen as valuable to supporting collaboration.                                                                          |
| Gender or sex                                                          | Two studies reported challenges with recruitment of people belonging to diverse                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                |

|                                               |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                       |
|-----------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| <i>(n=2 studies)</i>                          | gender or sexual identities in evidence synthesis research (Troya et al., 2019; Boote et al., 2011). Both articles reported concerns regarding recruitment, and in one article all patient/public members were white British heterosexual, and authors were unable to recruit members belonging to lesbian, gay, bisexual and transsexual (LGBT) groups (Troya et al., 2019).                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                         |
| Religion<br><i>(n=0 studies)</i>              | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                         |
| Education<br><i>(n=0 studies)</i>             | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                         |
| Socio-economic status<br><i>(n=3 studies)</i> | Three articles reported on issues related to access to technology, resulting in resource bias (Haddaway et al., 2017; Gavin et al., 2024; Wang et al., 2023). Review teams should address potential biases in engagement by minimizing the resources required for participation, offering multiple modes of interaction, and providing support for physical meeting attendance (Haddaway et al., 2017). While online engagement is generally positive, incorporating at least one in-person meeting per year can ensure inclusion of diverse voices and communities (Gavin et al., 2023), mitigating limitations of online-only recruitment, which may favor individuals with greater access and availability to the internet and technology (Wang et al., 2023).                                                                                                                                                                                                                     |
| Social capital                                | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                         |
| Age (plus)<br><i>(n=3 studies)</i>            | Three articles reported on the engagement of youth (Cooper et al., 2020; Walker et al., 2021; Smith et al., 2009). Empowering young voices in research is vital, especially as they are often underrepresented (Cooper et al., 2023) yet possess unique insights into issues regarding living with long-term health conditions (Walker et al., 2021). Involving interest-holders under 16 years old requires additional consent from caregivers, adding complexity to the process (Cooper et al., 2020). Ensuring discussions use age-appropriate language is crucial (Walker et al., 2021, while addressing self-doubt among participants is essential for inclusive engagement (Smith et al., 2009). Considering a blended co-production and consultation model can optimize involvement, allowing each individual to contribute their skills and expertise effectively (Walker et al., 2021).                                                                                      |
| Disability (plus)<br><i>(n=4 studies)</i>     | Four articles reported on facilitators to promote the engagement of people living with disability (Wang et al., 2023; Brutt et al., 2017; Walker et al., 2021; Boote et al., 2011). Facilitators for engagement include leveraging online platforms such as Facebook to connect with diverse interest-holders, for example closed groups such as “Patients Partnering in Research” (Wang et al., 2023). Workshops and tailored training sessions about evidence synthesis can enhance understanding of research processes, as seen in an initiative aimed at engaging patients with mental disorders (Brutt et al., 2017). Sensitivity to health conditions is crucial, requiring measures such as providing summaries and alternative feedback methods for those unable to attend meetings due to health issues (Walker et al., 2021). However, ensuring representativeness remains a challenge, as recruitment methods may not capture a truly diverse sample (Boote et al., 2011). |

## Principles to support equitable engagement

Our content analysis identified four key principles to support equitable engagement in evidence synthesis, which are summarized in Table 3 and expanded upon below.

**Table 3. Summary of principles to support equitable engagement**

| Principle                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                              | Contributing articles                                                                                                                                                                                                                                                                                                       |
|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| <b><i>Power-sharing and empowerment:</i></b> Empowerment involves providing resources to reduce power disparities and create conditions for broader decision-making, while power-sharing includes mechanisms to ensure diverse groups' inclusion in decision-making processes. The ultimate goal is to contribute to the empowerment of interest-holders, allowing them to set and achieve health goals, but challenges such as power imbalances and concerns about the independence of the review process need to be addressed through formal engagement methods which promote bi-directional partnerships.                                                                                                                                                                                                                                                           | Bastian et al., 2011<br>Boden et al., 2021<br>Boote et al., 2011<br>Boote et al., 2012<br>Buus et al., 2022<br>Cooper et al., 2020<br>Garrity et al., 2023<br>Harris et al., 2018<br>Langer et al., 2017<br>Morley et al., 2016<br>Pollock et al., 2022<br>Smith et al., 2009<br>Synnot et al., 2019<br>Walker et al., 2021 |
| <b><i>Accessibility and support:</i></b> Ensuring the accessibility of the evidence synthesis process for all interest-holders involves providing respectful and accessible training, including lay-friendly materials and clear information on the research purpose and methodology. Research activities, materials, and communication methods should be designed to accommodate diverse abilities and language preferences, with a focus on fostering open and accessible discussions. Maintaining flexibility in the approach throughout the synthesis process, addressing challenges like digital literacy, providing logistical support, and coordinating meetings at convenient times are crucial measures to meet the changing emotional and physical needs of interest-holders.                                                                                | Boden et al., 2021<br>Coon et al., 2016<br>Gavin et al., 2023<br>Pollock et al., 2022<br>Synnot et al., 2020<br>Troya et al., 2019<br>Walker et al., 2021                                                                                                                                                                   |
| <b><i>Diversity, intersectionality, and inclusion of marginalized groups (social justice):</i></b> Engaging diverse and intersectional interest-holders in evidence synthesis, including members of the research team, is seen as an opportunity for social justice, aiming to amplify historically disadvantaged voices and bring about sustained social change in policies, legislation, and public health outcomes. This inclusive approach is seen as essential for incorporating social factors that influence review findings. Efforts to ensure inclusivity and representativeness involve targeted recruitment, diversity among review authorship teams, and the use of culturally sensitive approaches to respect the differences among interest-holders, fostering trust and meaningful discussions through Equity, Diversity, and Inclusion (EDI) training. | Arnstein et al., 2020<br>Buus et al., 2022<br>Dewidar et al., 2022<br>Gribble et al., 2014<br>Haddaway et al., 2017<br>Harris et al., 2018<br>Nasser et al., 2013<br>Mogo et al., 2020<br>Morley et al., 2016<br>O'Connor et al., 2023<br>Synnot et al., 2020                                                               |
| <b><i>Ethical considerations:</i></b> Ethical challenges arise when including interest-holders' contributions about their lived experiences in evidence synthesis. While interest-holders with professional skills may not require ethical approval, the inclusion of lived experiences necessitates ethical considerations to avoid harm and manage                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                   | Abrams et al., 2021<br>Manafò et al., 2018<br>Pollock et al., 2022<br>Smith et al., 2009                                                                                                                                                                                                                                    |

|                                                                                                                                                                                                                                                                                                 |  |
|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--|
| risks. Addressing ethical concerns is crucial to prevent tokenistic engagement, ensure meaningful contributions, and involves realistic timelines, transparent communication about the use of contributions, and making the research process accessible and understandable to interest-holders. |  |
|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--|

## 1. Addressing power imbalances and promoting empowerment

Common identified principles for engagement were "empowerment" and "power-sharing", which involve the distribution and exercise of power within the evidence synthesis research process. Both terms aim to promote a more inclusive and equitable distribution of power. Empowerment involves providing resources and support to address barriers to engagement, reduce disparities in power, and create conditions where interest-holders contribute to broader decision-making processes (Smith et al., 2009; Harris et al., 2018). Power-sharing includes mechanisms such as proportional representation or arrangements to ensure the inclusion of different groups in decision-making processes throughout the evidence synthesis process. In this regard, "empowerment" was typically used to describe an approach to promote the engagement of patients and members of the public in evidence synthesis, whereas "power-sharing" typically addressed the relationship between patient/public partners and people with expertise in evidence synthesis methods (principal investigators), who traditionally hold decision-making authority over research processes.

The ultimate goal of engaging patients/public in evidence synthesis, particularly in prioritization or outcome selection, is to contribute to their empowerment and provide support for them to set and achieve their own health goals and ensure that the outcomes that matter to them are considered (Bastian et al., 2011; Harris et al., 2018; Morley et al., 2016). Theoretical frameworks for engagement and positive team dynamics can allow interest holders to present different points of view through robust discussion, while simultaneously seeing the impact of their participation on health research (Bastian et al., 2011; Boden et al., 2021). Empowering interest-holders' voices is particularly important for groups who are typically underrepresented within a literature base, such as youth (Cooper et al., 2020; Walker et al., 2021). Empowered patient and public groups can contribute to active engagement with broader evidence synthesis-



producing organizations, such as the Cochrane Collaboration (Morley et al., 2016). However, while some aspects of being engaged in evidence synthesis research might feel empowering to some interest-holders, others might feel disempowering (Smith et al., 2009). Case studies of engagement in systematic reviews have reported that clinical and university-based researchers voiced concerns about the power and authority of patient co-researchers due to a lack of trust (Boote et al., 2011). Reviews led by interest-holders demands may move the review design and conduct much closer to the user of the review. This approach changes the balance of power between the researcher and the review user, which can elicit worries about the independence of the review process and findings (Langer et al., 2017). This may result in the poor reporting of engagement exercises in evidence syntheses, which could indicate that some interest-holders are in a precarious position because of power imbalances between researchers and themselves (Buus et al., 2022).

Efforts should be made to mitigate potential power imbalances, such as a ‘leave your title at the door’ policy and encouraging activities that allow researchers and interest-holders to learn about each other outside of their roles on the team (Garritty, et al., 2023). Two studies suggested that formal engagement methods (e.g. nominal group techniques or Delphi methods) can help address concerns about group dynamics (Boote et al., 2012, Pollock et al., 2022), which can reduce the influence of overly dominant researchers while ensuring that the public’s contribution is considered alongside that of professionals (Boote et al., 2012). In one study among young people, youth leaders were recruited purposefully as facilitators for discussion to reduce the impact of perceived power imbalances between researchers and youth interest-holders (Cooper et al., 2020). In a second study, the investigators wanted at least half of their interest-holders to represent consumers (patients and the public) to mitigate potential power imbalances and ensure ample inclusion of non-professional perspectives (Synnot et al., 2019). Guidance from JBI outlines that it is important to promote and commit to bi-directional partnerships where different viewpoints are valued, there is co-benefit for all interest holders, power and responsibility are equally shared, and trust, transparency, and honesty are emphasized (Pollock et al., 2022).

## **2. Accessibility and support**

Working with interest-holders may necessitate additional measures to ensure that the evidence synthesis process is accessible to all. Lay summaries and accessible media can help engage excluded populations, but researchers must work to establish trusted partnerships in public communities (Gavin et al., 2023). Providing accessible and respectful training (capacity building) that familiarizes interest-holders with the research process, such as providing lay-friendly and accessible materials explaining the details of the research project, is a key process to establishing them as full research partners (Boden et al., 2021; Troya et al., 2019). Research activities, materials, and communication methods should be accessible to all interest-holders, including those with diverse abilities and language and cultural preferences. Orientation to the project should include clear and accessible information on the purpose of the evidence synthesis, how the synthesis will be carried out, and transparency about how perspectives from interest-holders will be incorporated (Walker et al., 2021). The research team should ensure that discussions are accessible and sensitive through the use of open and accessible language (Coon et al., 2016). Throughout the evidence synthesis process, research teams should maintain flexibility in their approach to accommodate changing emotional and physical needs and demands of interest holders (Troya et al., 2019; Walker et al., 2021). This may include addressing accessibility and challenges surrounding digital literacy (Pollock et al., 2022), providing logistical support (e.g., transportation costs and infrastructure) to ensure meeting venues are accessible (Troya et al., 2019; Pollock et al., 2022), as well as coordinating meetings at a date and time convenient for interest holders (Troya et al., 2019). Several articles also recommended being flexible and asking interest-holders to advise on how they would find it easiest to contribute to the project (e.g., by email, phone call before or after meeting, face-to-face, etc) (Coon et al., 2016; Pollock et al., 2022; Synnot et al., 2020; Walker et al., 2021).

### **3. Diversity, intersectionality, and inclusion of marginalized groups (social justice)**

The engagement of diverse and intersectional interest-holders was described as an opportunity for social justice, whereby engaging historically disadvantaged and silenced voices could promote knowledge transfer and create sustained social change (Buus et al., 2022), such as changes to policies, legislation, public health outcomes or clinical practice in favor of disadvantaged groups (Nasser et al., 2013; Mogo et al., 2020). Greater involvement of these

interest-holders in reviews can support greater inclusion of social factors that may influence review findings (Dewidar et al., 2022). Two articles argued that a stronger emphasis on social justice could transform evidence synthesis by adding a reflexive stance on the ‘politics’ of research (Buus et al., 2022; Gribble et al., 2014). Nasser and colleagues (2013) emphasized the proactive engagement of disadvantaged populations in their proposed approach to priority setting to promote equity in systematic reviews. Specifically, they outlined the engagement of different interest-holders who might be affected by the review topic (across PROGRESS characteristics) in the prioritization process and providing them with an opportunity to provide feedback and appeal the results of the prioritization (Nasser et al., 2013). However, participation in review processes is often low among Indigenous people and people from culturally and linguistically diverse backgrounds (Synnot et al., 2020). Indeed, engagement in review-producing organizations such as Cochrane is overwhelmingly Anglophone with a heavy concentration of consumer involvement in high income countries (Morley et al., 2016).

Despite these patterns, several articles recommended that inclusivity and representation should be pursued by working towards ensuring interest-holders reflect the population as a whole in respect of gender, age, ethnicity, disability, sexual orientation, socio-economic group, spoken language, and developed/developing world. This might be achieved through targeted recruitment programmes to improve the representation of currently under-represented interest-holders from low-income and non-English speaking countries (Morley et al., 2016). Arnstein and colleagues (2020) encouraged diversity among review authorship teams, which not only ensures that authors represent diverse interest-holder communities that can provide input from their lived experience, but also safeguards against limiting the definitions of outcomes and impacts used for the review (Harris et al., 2018). Using a broad, encompassing definition of “interest-holders” can help to ensure that all relevant groups are engaged, particularly minority groups and those in disadvantaged populations (Haddaway et al., 2017; O’Connor et al., 2023). To recognize and respect the differences in social identities of interest holders across PROGRESS-Plus characteristics, such as culture or language, evidence synthesis teams should embody culturally sensitive approaches and engage in ongoing efforts to build cultural competence among researchers and interest-holders in ways that promote co-learning. Taking Equity, Diversity, and Inclusion (EDI) training or participating in a self-reflective exercise together as a team can build

trust and foster a safe space for meaningful discussion (Dewidar et al., 2022; Garritty et al., 2023).

#### **4. Ethical considerations**

Inviting interest-holders to engage in key stages of an evidence synthesis respects both the expertise and the boundaries of that expertise (Abrams et al., 2021). It is, however, unclear how to do this in a way that is ethical, since evidence syntheses do not require ethics approval and there is no guidance on handling ethical dilemmas that may arise in using statements put forward by interest-holders (Abrams et al., 2021). For example, ethical approval to include interest-holders who are participating based on their professional skills, such as a librarian, methodologist, or another researcher, is usually not required as they are advising the research project from a professional capacity (Pollock et al., 2022). It is unclear whether or not it is acceptable to engage patients or members of the public in early research activities such as grant proposal or protocol writing before ethical approval has been obtained (Smith et al., 2009). Ethical approval may become particularly pertinent when including interest-holders' lived experiences, as it ensures that the risk of harm has been considered, discussed, and managed to avoid undue risk to the individual (Pollock et al., 2022). This may, however, introduce an unintended power dynamic between researchers and other interest-holders. In a review by Smith and colleagues (2009), the relationship between researchers and other interest-holders, intended to be a collaborative partnership, was contested by the ethics committee's requirement of informed consent and further compounded by the use of a consent form predicated on the signatory as a study subject (Smith et al., 2009), despite the engagement in reviews being considered low-risk (Pollock et al., 2022).

Furthermore, the risk of tokenism is high across all engagement opportunities (Manafò et al., 2018). Tokenistic engagement can occur when the relationship is only one-way; for example, the researcher includes interest-holders to acquire a grant, but then does not consult with them once funding is obtained (Pollock et al., 2022). Addressing ethical considerations is critical for avoiding inadequate allocation of time that may result in a lesser impact on public engagement, tokenism, and potentially leading interest-holders to being disenfranchised and unable to

contribute fully to the evidence synthesis (Manafò et al., 2018). Ways to embed an ethical perspective in engaged evidence synthesis include realistic timelines, transparency about how contributions are used in the research, and ensuring that the research process is accessible and communicated in a way that they can access and understand, using plain language (Manafò et al., 2018).

### **3.5 DISCUSSION**

Equitable engagement in health research infers a moral imperative to engaging interest-holders whereby people have a right to be involved in activities and decisions that may affect their health (Petkovic et al., 2020; Esmail et al., 2015). Reducing the known power imbalances between researchers and patients is a moral duty of researchers, especially with oppressed and seldom-heard groups (Greenhalgh et al., 2019). This scoping review sought evidence on equity considerations for the engagement of 11 different groups of interest-holders in the evidence synthesis process. We identified a small number of studies reporting the engagement of interest-holders across PROGRESS-Plus characteristics. Further, we summarized four key principles to support equitable engagement including power dynamics, accessibility, diversity and inclusion, and ethical considerations. The findings of this review complement a series of reviews by the MuSE Consortium, which will inform global guidance for the engagement of interest-holders in evidence synthesis (Tugwell et al., 2023). Other reviews in this series will summarize available guidance for and reporting of the engagement of interest-holders in evidence syntheses (Todhunter-Brown et al., 2024), barriers and facilitators to engagement (Campbell et al., in progress), impacts of engagement (Magwood et al., 2024), and conflict of interest considerations (Khabisa et al., in progress).

The engagement of interest-holders in research supports social justice, emphasizing a fair distribution of opportunities for people to achieve good health by addressing structural and systemic social disadvantage (Sen, 2002). Enabling individuals to exercise their agency— both individually and collectively —enables them to prioritize which health domains they value most and to choose what health services they would like to consume (Ruger, 2004). Opportunities for

individuals to exercise agency in health service and policy prioritization, design, delivery, or evaluation supports the growing movement for co-production in health. A central tenet of co-production in health is the empowerment of interest-holders to take control of their own health and health-related decisions (Loeffler et al., 2013). Co-production is characterized by mutual consultation and decision making between interest-holders and researchers, resulting in a sharing of knowledge between professionals and the communities affected by the health research, policy, or practice (Nowotny et al., 2013). In doing so, co-production may lead to improved health service quality, impact, and uptake while simultaneously reducing health and social inequities (Oliver et al., 2019). Notably, following a co-production approach may be beneficial to ensure interest-holder's autonomy in evidence synthesis by giving interest-holders the opportunity to choose how they want to be acknowledged in the research. For example, refugees who engage in evidence synthesis research may prefer to have their names excluded from publications in order to protect their safety and privacy, while ensuring that the perspectives of their lived experience are reflected in the evidence base.

However, the idea of bringing in non-traditional voices into health research has reportedly had very little influence on current evidence synthesis practices (Buus et al., 2022). Engaged evidence synthesis is dominated by high-income and English-speaking interest-holders, and very little is known about how interest holders from low-income settings, diverse cultures or languages, or different socio-economic positions could be engaged. Indeed, a recent systematic review was unable to identify guidance documents specifically addressing engaging racially or ethnically diverse interest-holders in evidence syntheses (Sathe et al, 2024). However, the authors did suggest that Indigenous research methods may provide models for equitable representation in syntheses and reporting of engagement. For example, a review by Morton Ninomiya and colleagues (2022) described practices for knowledge translation in the Indigenous health research field and was governed by Indigenous health scholars, Knowledge Guardians, and Elders. Such an approach could serve as an example of reflexive and equitable engagement for future evidence syntheses.

To truly address health inequities, health decision makers and researchers cannot afford to work in silos. Rather, they need to embrace co-produced health care and grow collaborative,

inclusive leadership across the public sector (Wilton, 2021). Future research should quantitatively evaluate the impact of engaging interest-holders in health decision making to ascertain the added value of such processes, and qualitatively explore the perspectives of interest-holders from an array of settings and research/policy domains to determine the most effective practices to support their engagement. Additionally, evidence synthesis teams will need to make pragmatic decisions in order for engagement to be feasible and worthwhile. Inherent value-based decisions will be needed in regards to how many interest-holders to include in an evidence synthesis initiative, their diversity across PROGRESS characteristics, and their representativeness. Further, the level of power-sharing across specific steps of the evidence synthesis process may require flexibility. For example, interest-holder engagement may be emphasized when setting the objectives and outcomes of the review, but may be less important in technical aspects of the review such as searching for evidence and conducting analyses (Björkqvist et al., 2021). However, such an approach risks being tokenistic and minimal, as evidence synthesis teams are not rewarded (either through publications or citations) for their engagement efforts. More research on methods, reporting and impact are needed to support evidence synthesis teams in their engagement approaches.

## **Strengths and limitations**

Our review is unique in that it addressed a knowledge gap around the equitable engagement of interest-holders in evidence synthesis (Tugwell et al., 2023). We searched a broad array of databases and gray literature sources to inform this scoping review and employed robust content analysis to characterize the equity considerations of engagement. However, several limitations should be noted. We primarily searched English-language databases, and it is possible that evidence from diverse linguistic groups or areas of the world may not have been identified by our search. Further, this review did not include individual systematic reviews and evidence syntheses on unrelated topics, as these will be included in a separate review being conducted by the MuSE Consortium (Todhunter-Brown et al., 2024). However, it is possible that these individual reviews contain further evidence on how equity and diversity is considered when engaging with interest-holders in evidence synthesis. Further, studies were only eligible if they

focused on evidence synthesis as a research method. This means that valuable findings from other research areas were omitted. For example, we anticipate overlap in the principles that support equitable engagement in evidence synthesis and best practices for community-based participatory research. Furthermore, in recent years several frameworks for engaged research have been developed (e.g., Corbie-Smith et al.; 2018; Jull et al., 2019; Hoekstra 2020), however none are specific to evidence synthesis and were therefore not considered for this review. We are also aware of resources which can support researchers in equitable engagement in research generally, such as the *Supporting equity-centred engagement* guide (Ul Haq et al., 2023), the *Equity in engagement framework* (HQO, 2020), and PCORI's *Equity and inclusion guiding engagement principles* (PCORI, 2021).

## Conclusions

Synthesized evidence in the form of evidence reviews (systematic reviews, scoping reviews, qualitative evidence synthesis, realist reviews) are the most common product to inform decision making for clinical practice, public health, and health systems (WHO et al., 2018). Increasing the capacity for health decision makers, academic research teams, and other interest-holders to work together on evidence syntheses can produce inclusive and actionable evidence that promotes the development of new health systems, practices, and different modes of knowledge and value production (Campbell & Vanderhoven, 2016; Felipe et al., 2017). The engagement of interest-holders in the synthesis of evidence may lead to modern health systems that ensure services reach the poorest, the most vulnerable, and those who are most often left behind (WHO et al., 2018). Evidence synthesis teams should emphasize sharing power amongst interest-holders, ensuring the evidence synthesis process is accessible to all, promoting diversity and inclusion, and integrating ethical considerations into their research practices. Future research should focus on tools to support evidence synthesis teams to conduct equitable engagement in research, including evaluative methods to assess impact.



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## Supplemental 1. PRISMA-ScR Checklist

| SECTION                   | ITEM | PRISMA-ScR CHECKLIST ITEM                                                                                                                                                                                                                                                 | REPORTED ON PAGE # |
|---------------------------|------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------|
| <b>TITLE</b>              |      |                                                                                                                                                                                                                                                                           |                    |
| Title                     | 1    | Identify the report as a scoping review.                                                                                                                                                                                                                                  | 25                 |
| <b>ABSTRACT</b>           |      |                                                                                                                                                                                                                                                                           |                    |
| Structured summary        | 2    | Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.                                             | 27-28              |
| <b>INTRODUCTION</b>       |      |                                                                                                                                                                                                                                                                           |                    |
| Rationale                 | 3    | Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.                                                                                                  | 28-30              |
| Objectives                | 4    | Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives. | 30                 |
| <b>METHODS</b>            |      |                                                                                                                                                                                                                                                                           |                    |
| Protocol and registration | 5    | Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.                                                                            | 30                 |
| Eligibility criteria      | 6    | Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.                                                                                                      | 31-33              |

|                                                       |    |                                                                                                                                                                                                                                                                                                            |               |
|-------------------------------------------------------|----|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------|
| Information sources*                                  | 7  | Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.                                                                                                  | 33            |
| Search                                                | 8  | Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.                                                                                                                                                                            | 67-80         |
| Selection of sources of evidence†                     | 9  | State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.                                                                                                                                                                                      | 33            |
| Data charting process‡                                | 10 | Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators. | 34            |
| Data items                                            | 11 | List and define all variables for which data were sought and any assumptions and simplifications made.                                                                                                                                                                                                     | 34            |
| Critical appraisal of individual sources of evidence§ | 12 | If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).                                                                                                      | N/A           |
| Synthesis of results                                  | 13 | Describe the methods of handling and summarizing the data that were charted.                                                                                                                                                                                                                               | 34            |
| <b>RESULTS</b>                                        |    |                                                                                                                                                                                                                                                                                                            |               |
| Selection of sources of evidence                      | 14 | Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.                                                                                                                               | 35            |
| Characteristics of sources of evidence                | 15 | For each source of evidence, present characteristics for which data were charted and provide the citations.                                                                                                                                                                                                | 35,36, 81-127 |

|                                               |    |                                                                                                                                                                                                 |        |
|-----------------------------------------------|----|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------|
| Critical appraisal within sources of evidence | 16 | If done, present data on critical appraisal of included sources of evidence (see item 12).                                                                                                      | N/A    |
| Results of individual sources of evidence     | 17 | For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.                                                           | 81-131 |
| Synthesis of results                          | 18 | Summarize and/or present the charting results as they relate to the review questions and objectives.                                                                                            | 37-48  |
| <b>DISCUSSION</b>                             |    |                                                                                                                                                                                                 |        |
| Summary of evidence                           | 19 | Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups. | 48-50  |
| Limitations                                   | 20 | Discuss the limitations of the scoping review process.                                                                                                                                          | 51     |
| Conclusions                                   | 21 | Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.                                       | 52     |
| <b>FUNDING</b>                                |    |                                                                                                                                                                                                 |        |
| Funding                                       | 22 | Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.                 | 27     |

## Supplemental 2. PRISMA-S Checklist

| Section/topic                          | # | Checklist item                                                                                                                                                                                                                                                     | Location(s) Reported |
|----------------------------------------|---|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------|
| <b>INFORMATION SOURCES AND METHODS</b> |   |                                                                                                                                                                                                                                                                    |                      |
| Database name                          | 1 | Name each individual database searched, stating the platform for each.                                                                                                                                                                                             | 33                   |
| Multi-database searching               | 2 | If databases were searched simultaneously on a single platform, state the name of the platform, listing all of the databases searched.                                                                                                                             | N/A                  |
| Study registries                       | 3 | List any study registries searched.                                                                                                                                                                                                                                | N/A                  |
| Online resources and browsing          | 4 | Describe any online or print source purposefully searched or browsed (e.g., tables of contents, print conference proceedings, web sites), and how this was done.                                                                                                   | 33                   |
| Citation searching                     | 5 | Indicate whether cited references or citing references were examined, and describe any methods used for locating cited/citing references (e.g., browsing reference lists, using a citation index, setting up email alerts for references citing included studies). | N/A                  |
| Contacts                               | 6 | Indicate whether additional studies or data were sought by contacting authors, experts, manufacturers, or others.                                                                                                                                                  | 33                   |
| Other methods                          | 7 | Describe any additional information sources or search methods used.                                                                                                                                                                                                | N/A                  |
| <b>SEARCH STRATEGIES</b>               |   |                                                                                                                                                                                                                                                                    |                      |
| Full search strategies                 | 8 | Include the search strategies for each database and information source, copied and pasted exactly as run.                                                                                                                                                          | 67-80                |

|                         |    |                                                                                                                                                                                           |       |
|-------------------------|----|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------|
| Limits and restrictions | 9  | Specify that no limits were used, or describe any limits or restrictions applied to a search (e.g., date or time period, language, study design) and provide justification for their use. | 67-80 |
| Search filters          | 10 | Indicate whether published search filters were used (as originally designed or modified), and if so, cite the filter(s) used.                                                             | 67-80 |
| Prior work              | 11 | Indicate when search strategies from other literature reviews were adapted or reused for a substantive part or all of the search, citing the previous review(s).                          | N/A   |
| Updates                 | 12 | Report the methods used to update the search(es) (e.g., rerunning searches, email alerts).                                                                                                | N/A   |
| Dates of searches       | 13 | For each search strategy, provide the date when the last search occurred.                                                                                                                 | 67-80 |
| <b>PEER REVIEW</b>      |    |                                                                                                                                                                                           |       |
| Peer review             | 14 | Describe any search peer review process.                                                                                                                                                  | 33    |
| <b>MANAGING RECORDS</b> |    |                                                                                                                                                                                           |       |
| Total Records           | 15 | Document the total number of records identified from each database and other information sources.                                                                                         | 67-80 |
| Deduplication           | 16 | Describe the processes and any software used to deduplicate records from multiple database searches and other information sources.                                                        | 33    |

### Supplemental 3. Search strategies

#### Ovid MEDLINE(R) ALL <1946 to May 19, 2022>

1 Stakeholder Participation/ 1988  
2 patient participation/ 28561  
3 consumer participation/ 18172  
4 Community-Based Participatory Research/ 5309  
5 Community-based participatory research.ti,ab,kf. 3636  
6 (coproduction or co-production).ti,ab,kf. 2836  
7 ((stakeholder\* or advisor\* or reference\* or expert\* or consultation\* or steering) adj2 (group\* or panel\*)).ti,ab,kf. 48206  
8 ((stakeholder\* or consultant\* or patient\* or consumer\* or public or caregiver\* or care-giver\* or communit\* or citizen\* or user\* or service-user\* or end-user\* or clinician\* or doctor\* or physician\* or nurse\* or policymaker\* or policy-maker\* or funder\* or indust\* or pharmac\* or payer\* or provider\* or pharmacist\*) adj2 (engag\* or involv\* or input or participat\* or collaborat\* or cooperat\*)).ti,ab,kf. 173373  
9 or/1-8 258670  
10 meta-analysis.pt. 160566  
11 meta-analysis/ or systematic review/ or meta-analysis as topic/ or "meta analysis (topic)"/ or "systematic review (topic)"/ or exp technology assessment, biomedical/ 302807  
12 ((systematic\$ adj3 (review\$ or overview\$)) or (methodologic\$ adj3 (review\$ or overview\$))).ti,ab. 267102  
13 ((quantitative adj3 (review\$ or overview\$ or synthes\$)) or (research adj3 (integrati\$ or overview\$))).ti,ab. 13669  
14 ((integrative adj3 (review\$ or overview\$)) or (collaborative adj3 (review\$ or overview\$)) or (pool\$ adj3 analy\$)).ti,ab. 34275  
15 (data synthes\$ or data extraction\$ or data abstraction\$).ti,ab. 35050  
16 (handsearch\$ or hand search\$).ti,ab. 10461  
17 (mantel haenszel or peto or der simonian or dersimonian or fixed effect\$ or latin square\$).ti,ab. 31850  
18 (met analy\$ or metanaly\$ or technology assessment\$ or HTA or HTAs or technology overview\$ or technology appraisal\$).ti,ab. 10285  
19 (meta regression\$ or metaregression\$).ti,ab. 12347  
20 (meta-analy\$ or metaanaly\$ or systematic review\$ or biomedical technology assessment\$ or bio-medical technology assessment\$).mp,hw. 405695  
21 (medline or cochrane or pubmed or medlars or embase or cinahl).ti,ab,hw. 294627  
22 (cochrane or (health adj2 technology assessment) or evidence report).jw. 20745  
23 (meta-analysis or systematic review).mp. 379456  
24 (comparative adj3 (efficacy or effectiveness)).ti,ab. 15291  
25 (outcomes research or relative effectiveness).ti,ab. 8386  
26 ((indirect or indirect treatment or mixed-treatment) adj comparison\$).ti,ab. 2521  
27 (meta-ethnograph\$ or metaethnograph\$ or meta ethnograph or meta-study or metastudy or meta study).ti,ab. 858

28 ((qualitative adj3 (review\$ or overview\$ or syntheses\$)) or (research adj3 (integrati\$ or overview\$))).ti,ab. 17202  
 29 (evidence adj2 map\$).ti,ab. 1217  
 30 ((evidence or realist) adj3 (review\$ or overview\$ or syntheses\$)).ti,ab. 76489  
 31 or/10-30 642113  
 32 9 and 31 21586

## CINAHL

S32, S8 AND S31 (11,454)

S31, S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15 OR S16 OR S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30 (301,653)

S30, TI (((evidence or realist) N2 (review\* or overview\* or syntheses\*))) OR AB (((evidence or realist) N2 (review\* or overview\* or syntheses\*))) (29,224)

S29, TI evidence N1 map\* OR AB evidence N1 map\* (437)

S28, TI (((qualitative N2 (review\* or overview\* or syntheses\*)) or (research N2 (integrati\* or overview\*)))) OR AB (((qualitative N2 (review\* or overview\* or syntheses\*)) or (research N2 (integrati\* or overview\*)))) (10,008)

S27, TI ((meta-ethnograph\* or metaethnograph\* or meta ethnograph or meta-study or metastudy or meta study)) OR AB ((meta-ethnograph\* or metaethnograph\* or meta ethnograph or meta-study or metastudy or meta study)) (23,996)

S26, TI (((indirect or indirect-treatment or mixed-treatment) W0 comparison\*)) OR AB (((indirect or indirect-treatment or mixed-treatment) W0 comparison\*)) (1,178)

S25, TI ("outcomes research" or "relative effectiveness") OR AB ("outcomes research" or "relative effectiveness") (3,942)

S24, TI (comparative N2 (efficacy or effectiveness)) OR AB (comparative N2 (efficacy or effectiveness)) (6,320)

S23, TI (meta-analys#s or (systematic w0 review\*)) OR AB ( meta-analys#s or (systematic w0 review\*)) OR MW ( meta-analys#s or (systematic w0 review\*)) (213,728)

S22, SO cochrane or (health\* N1 (technology W0 assessment\*)) or (evidence W0 report\*) (9,531)



S21, TI ((medline or cochrane or pubmed or medlars or embase or cinahl)) OR AB ((medline or cochrane or pubmed or medlars or embase or cinahl)) OR MW ((medline or cochrane or pubmed or medlars or embase or cinahl)) (160,626)

S20, TI (meta-analy\* or metaanaly\* or (systematic W0 review\*) or "biomedical technology assessment" or "biomedical technology assessments" or "bio-medical technology assessment" or "bio-medical technology assessments") OR AB (meta-analy\* or metaanaly\* or (systematic W0 review\*) or "biomedical technology assessment" or "biomedical technology assessments" or "bio-medical technology assessment" or "bio-medical technology assessments") OR MW (meta-analy\* or metaanaly\* or (systematic W0 review\*) or "biomedical technology assessment" or "biomedical technology assessments" or "bio-medical technology assessment" or "bio-medical technology assessments") (214,962)

S19, TI ((meta W0 regression\*) or metaregression\* ) OR AB ((meta W0 regression\*) or metaregression\*) (4,616)

S18, TI ((met W0 analy\*) or metanaly\* or (technology W0 assessment\*) or HTA or HTAs or (technology W0 overview\*) or (technology W0 appraisal\*)) OR AB ((met W0 analy\*) or metanaly\* or (technology W0 assessment\*) or HTA or HTAs or (technology W0 overview\*) or (technology W0 appraisal\*)) (4,623)

S17, TI ((mantel W0 haenszel) or peto or (der W0 simonian) or dersimonian or (fixed W0 effect\*) or (latin W0 square\*)) OR AB ((mantel W0 haenszel) or peto or (der W0 simonian) or dersimonian or (fixed W0 effect\*) or (latin W0 square\*)) (9,483)

S16, TI ( handsearch\* or hand-search\*) OR AB ( handsearch\* or hand-search\*) (4,892)

S15, TI ((data W0 synthes\*) or (data w0 extraction\*) or (data W0 abstraction\*)) OR AB ((data W0 synthes\*) or (data w0 extraction\*) or (data W0 abstraction\*)) (13,846)

S14, TI ((integrative N2 (review\* or overview\*) or (collaborative N2 (review\* or overview\*)) or (pool\* N2 analy\*)) OR AB ((integrative N2 (review\* or overview\*) or (collaborative N2 (review\* or overview\*)) or (pool\* N2 analy\*)) (16,854)

S13, TI ((quantitative N2 (review\* or overview\* or synthes\*) or (research N2 (integrati\* or overview\*))) OR AB ((quantitative N2 (review\* or overview\* or synthes\*) or (research N2 (integrati\* or overview\*))) (6,175)

S12, TI ((systematic\* N2 (review\* or overview\*) or (methodologic\* N2 (review\* or overview\*))) OR AB ((systematic\* N2 (review\* or overview\*) or (methodologic\* N2 (review\* or overview\*))) (137,240)

S11, MH Systematic Review (112,124)

S10, MH Meta Analysis (64,175)

S9, PT meta-analysis (4,226)

S8, S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 (116,320)

S7, TI ((stakeholder\* or consult\* or patient\* or consumer\* or client\* or public or caregiver\* or care-giver\* or communit\* or citizen\* or user\* or service-user\* or end-user\* or clinician\* or doctor\* or physician\* or nurs\* or policymaker\* or policy-maker\* or funder\* or indust\* or pharmac\* or payer\* or provider\* or carer or carers or healthcare\* or health-carer\* or healthprovider\* or health-provider\* or frontline\* or (lab\* N0 technician\*) or facult\* or frontliner# or front-liner#) N1 (engag\* or involv\* or input# or collaborat\* or cooperat\*)) OR AB ( (stakeholder\* or consult\* or patient\* or consumer\* or client\* or public or caregiver\* or care-giver\* or communit\* or citizen\* or user\* or service-user\* or end-user\* or clinician\* or doctor\* or physician\* or nurs\* or policymaker\* or policy-maker\* or funder\* or indust\* or pharmac\* or payer\* or provider\* or carer or carers or healthcare\* or health-carer\* or healthprovider\* or health-provider\* or frontline\* or (lab\* N0 technician\*) or facult\* or frontliner# or front-liner#) N1 (engag\* or involv\* or input# or collaborat\* or cooperat\*)) OR MW ((stakeholder\* or consult\* or patient\* or consumer\* or client\* or public or caregiver\* or care-giver\* or communit\* or citizen\* or user\* or service-user\* or end-user\* or clinician\* or doctor\* or physician\* or nurs\* or policymaker\* or policy-maker\* or funder\* or indust\* or pharmac\* or payer\* or provider\* or carer or carers or healthcare\* or health-carer\* or healthprovider\* or health-provider\* or frontline\* or (lab\* N0 technician\*) or facult\* or frontliner# or front-liner#) N1 (engag\* or involv\* or input# or collaborat\* or cooperat\*)) (69,794)

S6, TI ((stakeholder\* or advisor\* or reference\* or expert\* or steering) N1 (group\* or panel\* or organi#ation\* or body or lobb\* or part\*)) OR AB ((stakeholder\* or advisor\* or reference\* or expert\* or steering) N1 (group\* or panel\* or organi#ation\* or body or lobb\* or part\*)) OR MW ((stakeholder\* or advisor\* or reference\* or expert\* or steering) N1 (group\* or panel\* or organi#ation\* or body or lobb\* or part\*)) 26,428)

S5, TI (coproduction or co-production) OR AB (coproduction or co-production) OR MW (coproduction or co-production) (790)

S4, TI Community-based participatory research OR AB Community-based participatory research OR MW Community-based participatory research (2,029)

S3, MH Professional Practice, Research-Based OR MH Occupational Therapy Practice, Research-Based OR MH Physical Therapy Practice, Research-Based (2,255)

S2, MH Consumer Participation (23,024)

S1, MH Stakeholder Participation (2,020)

## SCOPUS

( TITLE-ABS ( "Community-based participatory research" OR ( coproduction OR co-production ) OR ( ( stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering ) W/1 ( group\* OR panel\* OR organi\*ation\* OR body OR lobb\* OR part\* ) ) OR ( ( stakeholder\* OR consult\*

OR patient\* OR consumer\* OR client\* OR public OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR frontliner\* OR front-liner\* ) W/1 ( engag\* OR involv\* OR input\* OR collaborat\* OR cooperat\* OR advice OR advise OR feedback ))) OR INDEXTERMS ( "Community-based participatory research" OR ( coproduction OR co-production ) OR ( ( stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering ) W/1 ( group\* OR panel\* OR organi\*ation\* OR body OR lobb\* ) ) OR ( ( stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR frontliner\* OR front-liner\* ) W/1 ( engag\* OR involv\* OR input\* OR collaborat\* OR cooperat\* ) ) ) ) AND ( ( TITLE-ABS ( ( ( systematic\* W/2 ( review\* OR overview\* ) ) OR ( methodologic\* W/2 ( review\* OR overview\* ) ) ) OR ( ( quantitative W/2 ( review\* OR overview\* OR synthes\* ) ) OR ( research W/2 ( integrati\* OR overview\* ) ) ) OR ( ( integrative W/2 ( review\* OR overview\* ) ) OR ( collaborative W/2 ( review\* OR overview\* ) ) OR ( pool\* W/2 analy\* ) ) OR ( data PRE/0 ( synthes\* OR extraction\* OR abstraction\* ) ) OR ( handsearch\* OR hand-search\* ) OR ( "mantel Haenszel" OR peto OR "der simonian" OR dersimonian OR ( fixed PRE/0 effect\* ) OR ( latin PRE/0 square\* ) ) OR ( met-analy\* OR metanaly\* OR hta OR htas OR ( technology PRE/0 ( overview\* OR appraisal\* OR assessment\* ) ) ) OR ( meta-regression\* OR metaregression\* ) OR ( comparative W/2 ( efficacy OR effectiveness ) ) OR ( "outcomes research" OR "relative effectiveness" ) OR ( ( indirect OR "indirect treatment" OR mixed-treatment ) PRE/0 comparison\* ) OR ( meta-ethnograph\* OR metaethnograph\* OR "meta ethnograph" OR meta-study OR metastudy OR "meta study" ) OR ( ( qualitative W/2 ( review\* OR overview\* OR synthes\* ) ) OR ( research W/2 ( integrati\* OR overview\* ) ) ) OR ( evidence W/1 map\* ) OR ( ( evidence OR realist ) W/2 ( review\* OR overview\* OR synthes\* ) ) ) ) OR ( TITLE-ABS-KEY ( ( meta-analy\* OR metaanaly\* OR ( systematic PRE/0 review\* ) OR ( biomedical PRE/0 technology PRE/0 assessment\* ) OR ( bio-medical PRE/0 technology PRE/0 assessment\* ) ) OR ( meta-analys\*s OR ( systematic PRE/0 review\* ) ) ) OR INDEXTERMS ( ( meta-analy\* OR metaanaly\* OR ( systematic PRE/0 review\* ) OR ( biomedical PRE/0 technology PRE/0 assessment\* ) OR ( bio-medical PRE/0 technology PRE/0 assessment\* ) ) ) ) OR ( SRCTYPE ( meta-analysis ) OR SRCTITLE ( cochrane OR ( health\* W/0 technology W/0 assessment\* ) OR ( evidence PRE/0 report\* ) ) ) OR ( TITLE-ABS ( ( medline OR cochrane OR pubmed OR medlars OR embase OR cinahl ) ) OR INDEXTERMS ( ( medline OR cochrane OR pubmed OR medlars OR embase OR cinahl ) ) ) )

## EMBASE

**Embase Classic+Embase <1947 to 2022 August 01>**

- 1 "patient participation"/ or "stakeholder engagement"/ or "participatory research"/44546
- 2 "community based participatory research".ti,ab. or ("consumer"/ and (participat\* or engag\*).ti,ab,kw.) 9722
- 3 (coproduction or co production).ti,ab,kw. 3302
- 4 ((stakeholder\* or advisor\* or reference\* or expert\* or consult\* or steering) adj2 (group\* or panel\* or organi?ation\* or body or lobb\* or part\*)).ti,ab,kw.
- 5 ((stakeholder\* or consult\* or patient\* or consumer\* or client\* or public or caregiver\* or 'care giver\*' or communit\* or citizen\* or user\* or 'service user\*' or 'end user\*' or enduser\* or clinician\* or doctor\* or physician\* or nurs\* or policymaker\* or 'policy maker\*') adj2 (engag\* or involv\* or input\$ or collaborat\* or cooperat\* or advice or advise or feedback)).ti,ab,kw. 197406

6 ((funder\* or indust\* or pharmac\* or payer\* or provider\* or carer or carers or healthcare\* or 'health carer\*' or healthprovider\* or 'health provider\*' or  
frontline\* or frontliner\$ or 'front liner\$') adj2 (engag\* or involv\* or input\$ or collaborat\* or cooperat\*)).ti,ab,kw. 23832

7 (('lab technician\*' or 'laboratory technician\*') adj2 (engag\* or involv\* or input\$ or collaborat\* or cooperat\*)).ti,ab,kw. 13

8 1 or 2 or 3 or 4 or 5 or 6 or 7 353983

9 ('meta analysis' or 'metaanalysis' or 'metanalysis').pt. 0

10 ((systematic\* adj3 (review\* or overview\*)) or (methodologic\* adj3 (review\* or overview\*))).ti,ab. 338290

11 ((quantitative adj3 (review\* or overview\* or synthes\*)) or (research adj3 (integrati\* or overview\*))).ti,ab. 16470

12 (((integrative adj3 (review\* or overview\*)) or (collaborative adj3 (review\* or overview\*)) or (pool\* adj3 analy\*))).ti,ab. 49644

13 ('data synthes\*' or 'data extraction\*' or 'data abstraction\*').ti,ab. 44001

14 (handsearch\* or 'hand search\*').ti,ab. 12883

15 ('mantel haenszel' or peto or 'der simonian' or dersimonian or 'fixed effect\*' or 'latin square\*').ti,ab. 43152

16 ('met analy\*' or metanaly\* or 'technology assessment\*' or hta or htas or 'technology overview\*' or 'technology appraisal\*').ti,ab. 17051

17 ('meta regression\*' or metaregression\*).ti,ab. 15666

18 ('meta analy\*' or metaanaly\* or 'systematic review\*' or 'biomedical technology assessment\*' or 'bio-medical technology assessment\*').ti,ab,de. 651032

19 (medline or cochrane or pubmed or medlars or embase or cinahl).ti,ab,de. 393428

20 (cochrane or (health\* adj2 'technology assessment\*') or 'evidence report\*').jt. 220

21 ('meta analys\$s' or 'systematic review\*').ti,ab,kw. 309697

22 (comparative adj3 (efficacy or effectiveness)).ti,ab. 23101

23 ('outcomes research' or 'relative effectiveness').ti,ab. 12992

24 ('indirect comparison\*' or 'indirect treatment comparison\*' or 'mixed treatment comparison\*').ti,ab. 5459

25 ('meta ethnograph\*' or metaethnograph\* or 'meta ethnograph' or metastudy or 'meta study').ti,ab. 995

26 ((qualitative adj3 (review\* or overview\* or synthes\*)) or (research adj3 (integrati\* or overview\*))).ti,ab. 20904

27 (evidence adj2 (map\$ or mapping)).ti,ab. 1412

28 ((evidence or realist) adj3 (review\* or overview\* or synthes\*)).ti,ab. 87785

29 exp meta analysis/ or "meta analysis (topic)"/ 299260

30 exp systematic review/ or "systematic review (topic)"/ 386923

31 'biomedical technology assessment'/ 15880

32 29 or 30 or 31 548234

33 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 32 926260

34 8 and 33 32355

## APA PSYCINFO

Set#: S1

Searched for: ("Stakeholder" AND (participat\* OR engag\*)) OR "Client Participation"

Databases: APA PsycInfo®

Results: 7158

Set#: S2

Searched for: ti(Community-based participatory research) OR ab(Community-based participatory research) OR if(Community-based participatory research)

Databases: APA PsycInfo®

Results: 2717

Set#: S3

Searched for: ti(coproduction or co-production) OR ab(coproduction or co-production) OR if(coproduction or co-production)

Databases: APA PsycInfo®

Results: 976

Set#: S4

Searched for: ti(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR ab(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR if(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*)))

Databases: APA PsycInfo®

Results: 47

Set#: S5

Searched for: ti(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback)) ) OR ab(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback)) ) OR mainsubject(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback)) )

Databases: APA PsycInfo®

Results: 130

Set#: S6

Searched for: (Systematic Review) OR (Meta Analysis)

Databases: APA PsycInfo®

Results: 97780

Set#: S7

Searched for: ti(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*)))) OR ab(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*))))

Databases: APA PsycInfo®

Results: 19

Set#: S8

Searched for: ti(((quantitative\* N2 (review\* OR overview\* OR syntheses\*) OR (research N2 (integrati\* OR overview\*)))) OR ab(((quantitative\* N2 (review\* OR overview\* OR syntheses\*) OR (research N2 (integrati\* OR overview\*))))

Databases: APA PsycInfo®

Results: 31

Set#: S9

Searched for: ti(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*))) OR ab(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*)))

Databases: APA PsycInfo®

Results: 9

Set#: S10

Searched for: ti((data W0 syntheses\*) OR (data W0 extraction\*) OR (data W0 abstraction\*)) OR ab((data W0 syntheses\*) OR (data W0 extraction\*) OR (data W0 abstraction\*))

Databases: APA PsycInfo®

Results: 0

Set#: S12

Searched for: ti(handsearch\* OR hand-search\*) OR ab(handsearch\* OR hand-search\*)

Databases: APA PsycInfo®

Results: 1485

Set#: S13

Searched for: ti((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*)) OR ab((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*))

Databases: APA PsycInfo®

Results: 163

Set#: S14

Searched for: ti((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs or (technology W0 overview\*) OR (technology W0 appraisal\*)) OR ab((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs or (technology W0 overview\*) OR (technology W0 appraisal\*))

Databases: APA PsycInfo®

Results: 426

Set#: S15

Searched for: ti((meta W0 regression\*) OR metaregression\*) OR ab((meta W0 regression\*) OR metaregression\*)

Databases: APA PsycInfo®

Results: 517

Set#: S16

Searched for: ti(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR ab(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR if(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments")

Databases: APA PsycInfo®

Results: 48469

Set#: S17

Searched for: ti((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR ab((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR if((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl))

Databases: APA PsycInfo®

Results: 32129

Set#: S18

Searched for: pub((cochrane OR (health\* N1 (technology W0 assessment\*)) OR (evidence W0 report\*)))

Databases: APA PsycInfo®

Results: 11

Set#: S19

Searched for: ti(meta-analyst#s OR (systematic W0 review\*)) OR ab(meta-analyst#s OR (systematic W0 review\*)) OR if(meta-analyst#s OR (systematic W0 review\*))

Databases: APA PsycInfo®

Results: 3

Set#: S20

Searched for: ti((comparative N2 (efficacy OR effectiveness))) OR ab((comparative N2 (efficacy OR effectiveness)))

Databases: APA PsycInfo®

Results: 1

Set#: S21

Searched for: ti("outcomes research" OR "relative effectiveness") OR ab("outcomes research" OR "relative effectiveness")

Databases: APA PsycInfo®

Results: 3805

Set#: S22

Searched for: ti(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*)) OR ab(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*))

Databases: APA PsycInfo®

Results: 0

Set#: S23

Searched for: ti((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study)) OR ab((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study))

Databases: APA PsycInfo®

Results: 40846

Set#: S24



Searched for: ti(((qualitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*)))) OR ab(((qualitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*))))

Databases: APA PsycInfo®

Results: 29

Set#: S25

Searched for: ti(evidence N1 map\*) OR ab(evidence N1 map\*)

Databases: APA PsycInfo®

Results: 17

Set#: S26

Searched for: ti(((evidence OR realist) N2 (review\* OR overview\* OR syntheses\*))) OR ab(((evidence OR realist) N2 (review\* OR overview\* OR syntheses\*)))

Databases: APA PsycInfo®

Results: 22

Set#: S27

Searched for: (("Stakeholder" AND (participat\* OR engag\*)) OR "Client Participation") OR (ti(Community-based participatory research) OR ab(Community-based participatory research) OR if(Community-based participatory research)) OR (ti(coproduction OR co-production) OR ab(coproduction OR co-production) OR if(coproduction OR co-production)) OR (ti(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR ab(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR if(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*)))) OR (ti(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback))) OR ab(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback))) OR mainsubject(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback))))

Databases: APA PsycInfo®

These databases are searched for part of your query.

Results: 10854

Set#: S28

Searched for: ((Systematic Review) OR (Meta Analysis)) OR (ti(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*)))) OR ab(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*)))) OR (ti(((quantitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*)))) OR ab(((quantitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*)))) OR (ti(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*))) OR ab(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*))) OR (ti((data W0 syntheses\*) OR (data W0 extraction\*) OR (data W0 abstraction\*)) OR ab((data W0 syntheses\*) OR (data W0 extraction\*) OR (data W0 abstraction\*)) OR (ti(handsearch\* OR hand-search\*) OR ab(handsearch\* OR hand-search\*)) OR (ti((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*)) OR ab((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*)) OR (ti((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs OR (technology W0 overview\*) OR (technology W0 appraisal\*)) OR ab((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs OR (technology W0 overview\*) OR (technology W0 appraisal\*))) OR (ti((meta W0 regression\*) OR metaregression\*) OR ab((meta W0 regression\*) OR metaregression\*)) OR (ti(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR ab(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR if(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments")) OR (ti((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR ab((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR if((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl))) OR pub((cochrane OR (health\* N1 (technology W0 assessment\*)) OR (evidence W0 report\*)) OR (ti(meta-analyst#s OR (systematic W0 review\*)) OR ab(meta-analyst#s OR (systematic W0 review\*)) OR if(meta-analyst#s OR (systematic W0 review\*))) OR (ti((comparative N2 (efficacy OR effectiveness))) OR ab((comparative N2 (efficacy OR effectiveness)))) OR (ti("outcomes research" OR "relative effectiveness") OR ab("outcomes research" OR "relative effectiveness")) OR (ti(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*)) OR ab(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*))) OR (ti((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study)) OR ab((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study))) OR (ti(((qualitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*)))) OR ab(((qualitative\* N2 (review\* OR overview\* OR syntheses\*)) OR (research N2 (integrati\* OR overview\*)))) OR (ti(evidence N1 map\*) OR ab(evidence N1 map\*)) OR (ti(((evidence OR realist) N2 (review\* OR overview\* OR syntheses\*)) OR ab(((evidence OR realist) N2 (review\* OR overview\* OR syntheses\*))))

Databases: APA PsycInfo®

These databases are searched for part of your query.

Results: 116400

Set#: S29

Searched for: (((("Stakeholder" AND (participat\* OR engag\*)) OR "Client Participation") OR (ti(Community-based participatory research) OR ab(Community-based participatory research) OR if(Community-based participatory research)) OR (ti(coproduction OR co-production) OR ab(coproduction OR co-production) OR if(coproduction OR co-production)) OR (ti(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR ab(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*))) OR if(((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consult\* OR steering\*) N1 (group\* OR panel\* OR organi#ation\* OR body OR lobb\* OR part\*)))) OR (ti(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback))) OR ab(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback))) OR mainsubject(((stakeholder\* OR consult\* OR patient\* OR consumer\* OR client\* OR public\* OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurs\* OR policymaker\* OR policy-maker\* OR funder\* OR indust\* OR pharmac\* OR payer\* OR provider\* OR carer\* OR carers OR healthcare\* OR health-carer\* OR healthprovider\* OR health-provider\* OR frontline\* OR (lab\* N0 technician\*) OR facult\* OR frontliner# OR front-liner#) N1 (engag\* OR involv\* OR input# OR collaborat\* OR cooperat\* OR advice OR advise OR feedback)))) AND (((Systematic Review) OR (Meta Analysis)) OR (ti(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*)))) OR ab(((systematic\* N2 (review\* OR overview\*)) OR (methodologic\* N2 (review\* OR overview\*)))))) OR (ti(((quantitative\* N2 (review\* OR overview\* OR synthes\*)) OR (research N2 (integrati\* OR overview\*)))) OR ab(((quantitative\* N2 (review\* OR overview\* OR synthes\*)) OR (research N2 (integrati\* OR overview\*)))))) OR (ti(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*))) OR ab(((integrative N2 (review\* OR overview\*)) OR (collaborative N2 (review\* OR overview\*)) OR (pool\* N2 analy\*)))) OR (ti((data W0 synthes\*) OR (data W0 extraction\*) OR (data W0 abstraction\*)) OR ab((data W0 synthes\*) OR (data W0 extraction\*) OR (data W0 abstraction\*))) OR (ti(handsearch\* OR hand-search\*) OR ab(handsearch\* OR hand-search\*)) OR (ti((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*)) OR ab((mantel W0 haenszel) OR peto OR (der W0 simonian) OR dersimonian OR (fixed W0 effect\*) OR (latin W0 square\*))) OR (ti((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs OR (technology W0 overview\*) OR (technology W0 appraisal\*)) OR ab((met W0 analy\*) OR metanaly\* OR (technology W0 assessment\*) OR HTA OR HTAs OR (technology W0 overview\*) OR (technology W0 appraisal\*))) OR (ti((meta W0 regression\*) OR metaregression\*) OR ab((meta W0 regression\*) OR metaregression\*)) OR (ti(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR ab(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments") OR if(meta-analy\* OR metaanaly\* OR (systematic W0 review\*) OR "biomedical technology assessment" OR "biomedical technology assessments" OR "bio-medical technology assessment" OR "bio-medical technology assessments")) OR (ti((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR ab((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR if((medline OR cochrane OR pubmed OR medlars OR embase OR cinahl)) OR pub((cochrane OR (health\* N1 (technology W0 assessment\*))) OR (evidence W0 report\*))) OR (ti(meta-analyst#s OR (systematic W0 review\*)) OR ab(meta-analyst#s OR (systematic W0

review\*)) OR if(meta-analyst#s OR (systematic W0 review\*)) OR (ti((comparative N2 (efficacy OR effectiveness))) OR ab((comparative N2 (efficacy OR effectiveness)))) OR (ti("outcomes research" OR "relative effectiveness") OR ab("outcomes research" OR "relative effectiveness")) OR (ti(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*)) OR ab(((indirect OR indirect-treatment OR mixed-treatment) W0 comparison\*)) OR (ti((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study)) OR ab((meta-ethnograph\* OR metaethnograph\* OR meta ethnograph OR meta-study OR metastudy OR meta study))) OR (ti(((qualitative\* N2 (review\* OR overview\* OR synthes\*)) OR (research N2 (integrati\* OR overview\*)))) OR ab(((qualitative\* N2 (review\* OR overview\* OR synthes\*)) OR (research N2 (integrati\* OR overview\*)))) OR (ti(evidence N1 map\*) OR ab(evidence N1 map\*)) OR (ti(((evidence OR realist) N2 (review\* OR overview\* OR synthes\*)) OR ab(((evidence OR realist) N2 (review\* OR overview\* OR synthes\*)))))

Databases: APA PsycInfo®

These databases are searched for part of your query.

Results: 546

# Supplemental 4: Characteristics of included studies table

| Study ID                                     | Study design                                                             | Objective                                                                                                                                                                                                                            | Type of evidence synthesis                                                                                                                                                                                                  | Definition of 'stakeholders'                                                                                                                                                                                                                                                                                                                                                                 | Definition of 'engagement'                                                                                                                                                                           | Results                      |        |                          |        |
|----------------------------------------------|--------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------|--------|--------------------------|--------|
|                                              |                                                                          |                                                                                                                                                                                                                                      |                                                                                                                                                                                                                             |                                                                                                                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                      | Framework or recommendations | Impact | Barriers or Facilitators | Equity |
| Abrams 2021<br><br>Companion:<br>Abrams 2020 | Review of realist reviews<br><br>n= 71 included studies<br><br>2014-2019 | The purpose of this review was to (1) describe the ways in which contributors have been involved in realist reviews, with a particular focus on patient/public involvement (PPI) and (2) document how involvement has been reported. | Realist reviews                                                                                                                                                                                                             | 'Contributors': non-researcher involvement including (but not limited to) stakeholders, patients and the public, decision and policy makers, experts and lay contributors.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers of care</li> <li>• Policymakers</li> <li>• Commissioners</li> <li>• Principal Investigators</li> </ul> | Consultations, informal stakeholder interviews, workshops "expert" or "advisory panel/groups"                                                                                                        | No                           | Yes    | No                       | No     |
| Agyei-Manu 2023                              | Systematic review<br><br>n=8 included studies                            | To synthesize qualitative evidence on the benefits, challenges, and best practices for Patient and Public Involvement (PPI) in evidence synthesis and systematic review projects from the                                            | Any evidence synthesis, also referred to as systematic review (SR), which seeks to answer a specific clinical or public health question by identifying, critically evaluating and synthesizing all the relevant evidence on | "Stakeholders" as defined by the 7Ps Framework: providers, policymakers, payers, purchasers, product makers, principal investigators (e.g., researchers) and patients/public.                                                                                                                                                                                                                | Patient and public involvement (PPI) describes the active process of effective partnership working with patients and members of the public to contribute to how health research is planned, managed, | Yes                          | Yes    | Yes                      | No     |

|               |                                                                        |                                                                                                                                                                                                                  |                                                                                                                                                                                                                                                                                          |                                                                                                                                                                                                                                                                                                                                               |                                                                                                                                                                                                                                                                                                         |     |     |     |     |
|---------------|------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|-----|-----|-----|
|               |                                                                        | perspectives of patients/public and researchers                                                                                                                                                                  | the topic.                                                                                                                                                                                                                                                                               | <p>This review focuses on patients/public, defined as: potential patients, carers and people who use health and social care services, people from organizations that represent people who use services</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> </ul>                                      | designed, conducted and disseminated. PPI denotes 'research being carried out "with" or "by" members of the public, rather than "to", "about" or "for" them'.                                                                                                                                           |     |     |     |     |
| Anderson 2021 | <p>Series of case studies</p> <p>n=8</p> <p>2017-2020</p> <p>UK</p>    | To describe and discuss the experiences of review scoping of three commissioned research centres that conducted evidence syntheses to inform health and social care organisation, delivery and policy in the UK. | Any evidence synthesis approach (rapid, scoping, realist, systematic, mapping): Specifically the planning stages between the selection of a review topic and the final review protocol (including final review questions, inclusion criteria and the type of planned evidence synthesis) | <p>Stakeholders: those who may use or be affected by the outcomes of research.. Referred to as "stakeholders", "end-users", "customers" and "service users".</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Policymakers</li> <li>• Commissioners</li> <li>• Payers of Research</li> </ul> | Co-production: The process whereby stakeholders work as partners with researchers to create, shape and deliver syntheses of research evidence. Can refer to the relationship between researchers and the potential end-users of the service, and the relationship between the funder and research team. | Yes | Yes | Yes | No  |
| Arnstein 2020 | <p>Systematic review</p> <p>n= 9 included studies</p> <p>2015-2017</p> | To (1) conduct a systematic review of the evidence on patient involvement in results sharing, (2) propose evidence-based recommendations to                                                                      | Systematic reviews and other peer-reviewed publications                                                                                                                                                                                                                                  | 'Patient' refers to "people having or at risk of having medical condition(s), whether or not they currently receive medicines or vaccines to prevent or treat a disease" as well                                                                                                                                                              | Involvement in the sharing of health research studies engaged as authors or contributors.                                                                                                                                                                                                               | Yes | Yes | Yes | Yes |

|               |                                                                                                                           |                                                                                                                                                                                |                                                                                                                                                                                                  |                                                                                                                                                                                                                                  |                                                                                |     |     |     |     |
|---------------|---------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------|-----|-----|-----|-----|
|               |                                                                                                                           | help maximize benefits and minimize risks of such involvement and (3) conduct this project with patient authors                                                                |                                                                                                                                                                                                  | as “the family and those voluntarily caring for those with the medical condition(s), patient advocates and patient groups.”<br><br>Groups:<br><ul style="list-style-type: none"> <li>Patients and caregivers</li> </ul>          |                                                                                |     |     |     |     |
| Aschmann 2020 | Case study<br><br>A project on informing care in people with multiple chronic conditions<br><br>Setting/year not reported | To glean important aspects for patient-centeredness in quantitative benefit-harm assessments (BHAs) and describe approaches we developed to this purpose                       | Quantitative benefit-harm assessments (BHAs) -- systematic, comprehensive assessments that integrate the evidence on all relevant outcomes and all key determinants of the benefit-harm balance. | Not defined, referred to as “stakeholder representatives”.<br><br>Groups:<br><ul style="list-style-type: none"> <li>Patients and caregivers</li> <li>Providers of care</li> <li>Commissioners [guideline developers]</li> </ul>  | Not defined. Referred to as “co-investigators”                                 | Yes | Yes | No  | No  |
| Bastian 2011  | Case study<br><br>Institute for Quality and Efficiency in Health Care (IQWiG), Germany<br><br>2006-2010                   | To establish criteria for assessing interest to and for patients/consumers, and to develop methods for choosing which systematic review results to summarize for the community | Systematic reviews or health technology assessments (HTAs)                                                                                                                                       | Not defined, referred to as “stakeholders”.<br><br>Groups:<br><ul style="list-style-type: none"> <li>Patients</li> <li>Public</li> <li>Providers of care</li> <li>Policymakers</li> <li>Peer reviewed journal editors</li> </ul> | Not defined. Referred to as “involvement in priority setting”                  | Yes | No  | No  | Yes |
| Bayliss 2016  | Cross-sectional impact evaluation<br><br>n=8 patients,                                                                    | To evaluate the patient experience of involvement in a qualitative meta-synthesis                                                                                              | Qualitative meta-synthesis                                                                                                                                                                       | ‘Patient research partners’ (PRPs), included People with Arthritis/Rheumatism in Europe (PARE) board                                                                                                                             | Not defined. Referred to as “research partners” and “involvement” in analysis. | Yes | Yes | Yes | Yes |

|                   |                                                                             |                                                                                                                                                                                                                                                                                                                                                                                                             |                                      |                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                                                                                                                                                                             |     |     |     |     |
|-------------------|-----------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|-----|-----|-----|
|                   | Europe (UK, Sweden, Estonia, Romania)<br><br>2014-2015                      |                                                                                                                                                                                                                                                                                                                                                                                                             |                                      | members/founders and members of national rheumatic and RA patient organizations<br><br>Groups:<br>● Patients                                                                                                                                                                        |                                                                                                                                                                                                                                                                                                                                                                             |     |     |     |     |
| Boden 2021        | Rapid review<br><br>n=9 included studies<br><br>1998-2019                   | To better understand best practices in engaging patients in synthesis reviews, answering two questions: (1) What are the characteristics of a review team that create an 'interactive team and positive research environment that is conducive to effective contributions by patients'? (2).How can research teams conduct their reviews to ensure 'planned, supported and valued involvement of patients'? | Systematic reviews and rapid reviews | "Patient partners", referring to patients as "an overarching term that includes individuals with personal experience of a health issue and informal caregivers, including family and friends"<br><br>Groups:<br>● Patients and caregivers<br>● Principal investigators (librarians) | "Meaningful engagement" in systematic reviews, defined as "the planned, supported and valued involvement of patients in the research process within an interactive team and positive research environment that facilitates effective contributions by patients or their surrogates to help to produce important outcomes while benefiting the patients or their surrogates" | Yes | Yes | Yes | Yes |
| Boote 2011 & 2012 | Systematic review with narrative synthesis (2011) with update and expansion | To review the evidence on public involvement in the systematic review process in health and social care and how relevant organizations                                                                                                                                                                                                                                                                      | Systematic reviews                   | 'The public' defined as, "patients and potential patients; people who use health and social services; informal carers; parents/guardians;                                                                                                                                           | 'Public involvement': "doing research 'with' or 'by' the public, rather than 'to', 'about' or 'for' the public". Three main levels: (1) consultation; (2) collaboration; (3)                                                                                                                                                                                                | No  | Yes | Yes | Yes |



|              |                                                                                                              |                                                                                                                                                                                                                                                                              |                                                                       |                                                                                                                                                                                                                                                                                                                                          |                                                                                                                                                                                                                                                                                                                                                                                                                                                                   |    |     |     |    |
|--------------|--------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----|-----|-----|----|
|              | <p>(2012)</p> <p>n=16 included studies (8 individual SRs and 9 organizational surveys)</p> <p>Up to 2011</p> | involve the public in their review processes; to examine the different methods, levels and stages of involving the public; to synthesize the contributions of the public, as well as the identified tensions, facilitating strategies and recommendations for good practice. |                                                                       | <p>disabled people; members of the public who are potential recipients of health promotion programmes, public health programmes and social service interventions; organizations that represent people who use services"</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients and caregivers</li> <li>• Public</li> </ul> | <p>'publicly led'.</p> <p>Public involvement in systematic reviews: Involvement in one or more of the following review activities : (1) review question; (2) review protocol (including search strategy and inclusion/exclusion criteria); (3) literature search; (4) appraisal of the literature including assessment of study relevance, data extraction and evidence synthesis; (5) drafting of findings; (6) interpretation of findings; (7) final report</p> |    |     |     |    |
| Bragge 2011  | <p>Case study of an evidence map</p> <p>Australia</p> <p>2007 onwards</p>                                    | To describe the Global Evidence Mapping (GEM) Initiative's evidence mapping methods and illustrate how these methods were applied to neurotrauma topics.                                                                                                                     | <p>Evidence maps</p> <p>Scoping reviews</p> <p>Systematic reviews</p> | <p>"Key stakeholders" described as patients, carers, clinicians, researchers and policymakers.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients and caregivers</li> <li>• Providers of care</li> <li>• Principal investigators</li> <li>• Policymakers</li> </ul>                                                    | Described as "expert consultation", including unstructured discussion, participation in workshops using the Nominal Group Technique, and online surveys.                                                                                                                                                                                                                                                                                                          | No | No  | No  | No |
| Brunton 2020 | <p>Systematic review</p>                                                                                     | To examine what is known about                                                                                                                                                                                                                                               | Systematic reviews using a framework                                  | Described as 'stakeholders', including                                                                                                                                                                                                                                                                                                   | Described as "involvement" (no                                                                                                                                                                                                                                                                                                                                                                                                                                    | No | Yes | Yes | No |

|              |                                                                                                     |                                                                                                                                                                                                                                                                                                      |                    |                                                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                                                                                                                                                                                         |     |     |    |     |
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|              | n= 61 included studies<br><br>Up to 2019                                                            | framework analysis methods in health research synthesis by addressing three questions: (1) How do methods of framework synthesis vary? (2) Where is framework synthesis located within the context of research synthesis methods? (3) What are the strengths and limitations of framework synthesis? | synthesis          | patients, caregivers, healthcare professionals, public<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients and caregivers</li> <li>• Public</li> <li>• Providers of care</li> </ul>                                                                                                                                                                               | definition).                                                                                                                                                                                                                                                                                            |     |     |    |     |
| Brütt 2017   | Mixed methods (focus groups and quantitative evaluation)<br><br>N = 7 patients, Germany<br><br>2015 | The aims of the present study were a) that patients contribute and prioritize clinically relevant outcomes of meta-cognitive interventions, and b) that they evaluate a patient workshop and give their perceptions of research involvement.                                                         | Systematic reviews | Distinguishes 'public involvement', which refers to involvement of members of the public or via patient groups or organizations, from patient involvement (PI) as the participation of users or patients based on their individual experience with a disease or with specific health interventions<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> </ul> | 'Involved' in health research implies that patients act as partners with valuable expertise, perspectives, and insights.<br><br>Patient involvement in systematic reviews focuses on identifying and prioritizing topics, developing the protocol, conducting the review, and disseminating the results | No  | Yes | No | Yes |
| Buckley 2014 | Case study using 18 structured discussions                                                          | To describe various Agency for Healthcare Research and Quality Evidence-based Practice                                                                                                                                                                                                               | Systematic reviews | 'Stakeholders' are those who would make decisions with the report findings, as well as those who would be                                                                                                                                                                                                                                                                         | Involving stakeholders in the nomination process includes providing an opportunity for end                                                                                                                                                                                                              | Yes | No  | No | No  |

|            |                                                                                                                                     |                                                                                                                                                                                                                                                                                       |                                                   |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                               |                                                                                                                                                                                                                                                       |    |     |    |    |
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|            | Agency for Healthcare Research and Quality Evidence-based Practice Center (EPC) Program, USA<br><br>2007                            | Center (EPC) Program approaches to the refinement process, with their advantages and disadvantages, and provides guidance for a consistent approach to refinement and the reporting of refinement decisions in light of guiding principles                                            |                                                   | <p>affected by those decisions. Stakeholders may represent patients, consumers, advocacy organizations, clinicians, researchers, agencies that issue guidelines, policy makers, industry, or health-care organizations.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients and caregivers</li> <li>• Providers of care</li> <li>• Principal investigators</li> <li>• Commissioners</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Program Managers</li> </ul> | users of research to participate in asking and answering questions about health care. Specifically, engaging them in "key informant interviews" or "public comment" opportunities.                                                                    |    |     |    |    |
| Burns 2018 | Case study of a Health Technology Assessment on reinforced home-based palliative care.<br><br>INTEGRATE-HTA Project, European Union | 1) Describe the scope of a systematic review of effectiveness, 2) Describe the combination of methods applied for summarizing, presenting, and further exploring the evidence included in this review, which included creating harvest plots and a subsequent gap analysis and expert | Systematic review, harvest plots and gap analysis | <p>'Stakeholders' included palliative care practitioners and researchers</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers of care</li> <li>• Principal investigators</li> </ul>                                                                                                                                                                                                                                                                                          | 'Consultation': Each expert was provided the opportunity to study the review protocol and the harvest plots and was asked to discuss methodological or palliative care related issues relevant to the emerging aspects arising from the gap analysis. | No | Yes | No | No |

|           |                                                                                                        |                                                                                                                                                                                                                                                                                                                                                                |                 |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                             |    |    |    |     |
|-----------|--------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----|----|----|-----|
|           |                                                                                                        | consultations. 3) Present the results from this combined approach to evidence synthesis and interpretation and briefly discuss the strengths and limitations of the applied methods.                                                                                                                                                                           |                 |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                             |    |    |    |     |
| Buus 2022 | <p>Critical literature review</p> <p>n= 66 included studies (convenience sample)</p> <p>Up to 2020</p> | <p>To explore how consultation exercises were described in a convenience sample of recent scoping reviews. Guided by three research questions: (1) At what stage of scoping studies were stakeholders consulted? (2) Who were the stakeholders and how were they consulted? and (3) How was information from stakeholders integrated into scoping studies?</p> | Scoping reviews | <p>'Stakeholder consultants' have experience in the field and were named 'stakeholders', 'users by experience', 'experts', 'clinicians', 'topic population', 'academic experts', 'expert key informants'. The stakeholder consultants had different roles, such as professionals, researchers, consumer representatives and organizational decision-makers and leaders.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers of care</li> <li>• Principal investigators</li> <li>• Patients and caregivers</li> <li>• Public</li> <li>• Policymakers</li> <li>• Program</li> </ul> | 'Consultation' is conducted individually (e.g., interviews and surveys) or in groups (e.g., focus groups, workshops, public discussion, expert panels and advisory groups). | No | No | No | Yes |

|                             |                                                                                                                                                                                                             |                                                                                                                                                                                                                                                        |                             | Managers                                                                                                                                                                                                                                                                                                                                                                                                                                                                      |                                                                                                                                                                                                                                                                                                                                                                                                                         |     |    |     |     |
|-----------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|----|-----|-----|
| Cochrane Collaboration 2015 | <p>Mixed method evaluation, Gray literature report</p> <p>Surveys (2014-2015) and literature review (1990-2015)</p> <p>n=42 review groups, 117 members of the Cochrane Consumer Network (international)</p> | To evaluate how well the current functions and structures of the Consumer Network support the strategic goals of Cochrane and to consider what changes are needed to ensure the organization has the right structures in place to achieve its mission. | Cochrane systematic reviews | <p>Consumers refers to patients; carers and family members; people who work paid or unpaid for an organization that supports the aims of Cochrane (for example a not for profit organization, patient-led or patient advocacy group); and less often, healthcare professionals; researchers and the public.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers of care</li> <li>• Principal investigators</li> <li>• Public</li> </ul> | <p>“Involvement” in reviews throughout the research cycle: from priority setting; outcomes identification; commenting on protocols reviews and plain language summaries; helping to disseminate knowledge and playing important roles within Review Group teams. It is also clear from the literature that consumer involvement historically has been focused on commenting on reviews and plain language summaries</p> | No  | No | Yes | Yes |
| Cochrane Collaboration 2022 | <p>Guidance, Gray literature report</p> <p>Strategic framework for Cochrane (international)</p>                                                                                                             | To describe a framework whose vision is to engage with healthcare consumers (patients, carers, and the public) and those who support them, so that they can make sense of health evidence, be integral to its production and dissemination, and        | Cochrane systematic reviews | <p>“Consumers” are made up from a wide range of people, including patients (or people with personal experience of a healthcare condition), health and social care service users, carers and family members.</p> <p>“Stakeholders” can be defined as any potential knowledge user whose</p>                                                                                                                                                                                    | <p>Involvement in research, defined as “research being carried out ‘with’ or ‘by’ members of the public rather than ‘to’, ‘about’ or ‘for’ them”. This differs from engagement which might include raising awareness of research (Cochrane evidence for example), sharing</p>                                                                                                                                           | Yes | No | No  | Yes |

|                |                                                                                                                                                 |                                                                                                                                          |                                      |                                                                                                                                                                                                                                                                                                                                                                                                                                         |                                                                                                                 |     |    |     |    |
|----------------|-------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------|-----|----|-----|----|
|                |                                                                                                                                                 | use Cochrane evidence when making healthcare decisions.                                                                                  |                                      | <p>primary job is not directly in research and can include people with experience of a health condition, their caregivers, families and patient and charitable support organizations. Wider stakeholders may include health and social care providers, policy makers and others.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers of care</li> <li>• Policymakers</li> <li>• Public</li> </ul> | knowledge or engaging and creating a dialogue with the public                                                   |     |    |     |    |
| Collins 2019   | <p>Commentary</p> <p>Authors experiences of producing systematic reviews with Government Departments and Agencies within the UK and Europe,</p> | To introduce the challenges of engaging policy-makers with systematic reviews, identify possible solutions and share our lessons learned | Systematic reviews                   | <p>Refers to “stakeholders” and specifically focuses on policymakers.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> </ul>                                                                                                                                                                                                                                                                               | “Engagement” without further definition.                                                                        | No  | No | Yes | No |
| Concannon 2014 | <p>Systematic review</p> <p>n= 95 included studies</p>                                                                                          | To catalog reported methods of stakeholder engagement in comparative effectiveness                                                       | Systematic reviews and meta-analyses | ‘Stakeholder’: An individual or group who is responsible for or affected by health-and healthcare-related decisions that can be                                                                                                                                                                                                                                                                                                         | Engagement: A bi-directional relationship between stakeholder and researcher that results in informed decision- | Yes | No | Yes | No |

|           |                                                                                                                     |                                                                                                                                                                                                                                                                                    |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                                                                     |     |     |     |    |
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|           | 2003-2012<br>USA                                                                                                    | research and patient-centered outcomes research, answering the following research question: How have U.S. researchers reported engagement with stakeholders in the prioritization, conduct and use of comparative effectiveness and patient-centered outcomes research since 2003? |                    | <p>informed by research evidence. The 7Ps Framework suggests that stakeholders may be organized into seven types:</p> <ul style="list-style-type: none"> <li>• Patients and the public,</li> <li>• Providers</li> <li>• Purchasers</li> <li>• Payers</li> <li>• Policy makers</li> <li>• Product makers</li> <li>• Principal investigators</li> </ul>                                                                                                               | making about the prioritization, conduct and use of research.                                                                                                                                                                                                       |     |     |     |    |
| Coon 2016 | <p>Systematic review with end-user involvement. This paper describes the experiences of team members.</p> <p>UK</p> | To (i) highlight the methods of end-user involvement used in our reviews, (ii) to facilitate and stimulate discussion of the most appropriate and efficient methods of engagement and (iii) to develop a list of suggestions to improve future involvement in systematic reviews.  | Systematic reviews | <p>'End users', described as "parents of children with ADHD, special educational needs specialists and experts in developing and delivering non-pharmacological interventions for ADHD in the school setting". "Expert Advisors" included individuals from an academic perspective, a charity perspective, and an intervention perspective (developer of an intervention).</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers of care</li> </ul> | <p>'Involvement' at two levels:</p> <p>1) Collaborative level: End users are an integral part of the project team</p> <p>2) Consultative level: individuals commenting on the protocol, editing draft chapters and responding to ad hoc questions as they arose</p> | Yes | Yes | Yes | No |

|              |                                                                                                                                                                    |                                                                                                                                                                                                                     |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                    |                                                                                                                                                                                                                                                                                                                                     |     |     |     |     |
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|              |                                                                                                                                                                    |                                                                                                                                                                                                                     |                    | <ul style="list-style-type: none"> <li>Principal investigators</li> <li>Patients and caregivers</li> </ul>                                                                                                                                                                                                                                                                                                                                         |                                                                                                                                                                                                                                                                                                                                     |     |     |     |     |
| Cooper 2020  | <p>Realist review (evaluative synthesis) with interviews and focus groups. This paper describes the experiences of the team members</p> <p>1980-2016</p> <p>UK</p> | To set out the challenges faced in conducting a realist review, outline the steps taken to customize the realist methodology, and to discuss how this customized methodology was used to overcome these challenges. | Realist reviews    | <p>'Stakeholders' included those involved in the development and delivery of adolescent risk behavior prevention programs, such as researchers, teachers, community youth workers, Personal, Social, Health and Economic education (PSHE) leaders, and peer coordinators.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>Providers of care</li> <li>Principal investigators</li> <li>Patients and caregivers</li> <li>Public</li> </ul> | 'Engagement' described as "meaningful participation", which goes beyond the informative capacity of asking stakeholders for advice moving more towards cooperation and collaboration in which stakeholder evidence has direct and clear impact on research outcomes which is clearly demonstrated in any relevant outputs produced. | No  | Yes | Yes | Yes |
| Cornman 2017 | <p>Interviews and focus group discussions to assess satisfaction, experiences and perceptions of being a key informant.</p> <p>N = 6 caregivers and</p>            | To examine how patients, caregivers, and patient advocates who participated as Key Informants in prior systematic reviews regarded that experience and what their recommendations are for improving that process.   | Systematic reviews | "Key Informants" are selected for a panel that includes a broad range of health care stakeholders such as clinicians, prominent researchers in the area, and individuals who can represent the patient perspective, such as patients, caregivers, and patient advocates                                                                                                                                                                            | The inclusion of the patient perspective in Topic Refinement of a systematic review                                                                                                                                                                                                                                                 | Yes | Yes | Yes | No  |



|               |                                                                                                                                                                                                                                               |                                                                                                                                 |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |                                                         |    |     |     |    |
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|               | <p>patient advocates</p> <p>USA, 2016-2017</p>                                                                                                                                                                                                |                                                                                                                                 |                    | <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients and caregivers</li> <li>• Providers</li> <li>• Principal investigators</li> </ul>                                                                                                                                                                                                                                                                                                                                                                                                                         |                                                         |    |     |     |    |
| Cottrell 2014 | <p>Literature review and interviews with program leaders, systematic reviewers and stakeholders from evidence-based programs.</p> <p>n=24 included studies and 34 participants in 12 focus groups discussions</p> <p>2004-2013</p> <p>USA</p> | To examine the benefits and challenges of engaging stakeholders in the process of developing and performing systematic reviews, | Systematic reviews | <p>“Stakeholder”, defined as a person or group with a vested interest in a particular clinical decision and the evidence that supports that decision, including:</p> <ul style="list-style-type: none"> <li>• Patients, caregivers, and patient advocacy organizations</li> <li>• Clinicians and professional associations</li> <li>• Institutional health care providers</li> <li>• Government agencies</li> <li>• Purchasers and payers</li> <li>• Health care industry</li> <li>• Health care policymakers</li> <li>• Researchers and research institutions.</li> </ul> | Referred to as “engagement” without further definition. | No | Yes | Yes | No |
| Cumpston 2012 | Interviews                                                                                                                                                                                                                                    | To identify policy makers’ priority                                                                                             | Systematic reviews | <p>Policymaker representatives from</p>                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                    | ‘Engagement’, described as                              | No | No  | No  | No |

|              |                                                                                                                                                              |                                                                                                                                                                                                                                                                 |                                                         |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                              |                                                                              |           |            |           |            |
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|              | <p>n=30 policymakers from the State and Territory Health Departments</p> <p>July to October 2008, Australia</p>                                              | <p>topics for which systematic reviews of the available evidence would be useful; to explore the extent to which existing Cochrane systematic reviews are relevant to these policy priorities; and to identify gaps where new systematic reviews are needed</p> |                                                         | <p>health jurisdiction. Their responsibilities included quality and safety, workforce, health system performance, rural health, primary and community care, Indigenous health, public health, and other areas.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> </ul>                                                                                                                                                                                                                           | <p>communication and collaboration between policymakers and researchers.</p> |           |            |           |            |
| Dennett 2022 | <p>Descriptive case study of an online survey prioritization process</p> <p>23 July to the 9 September 2019, international</p> <p>n=147 survey responses</p> | <p>To prioritize 10 reviews of importance to the public (patients, carers, healthcare professionals and researchers) from a patient survey and develop systematic review questions.</p>                                                                         | <p>Priority setting for Cochrane systematic reviews</p> | <p>“Stakeholders” were people living with a respiratory condition or carers of someone with a respiratory condition, Cochrane or external stakeholders such as healthcare professionals, including doctors and nurses, people linked to organizations that are involved with healthcare research, providing patient information, developing guidelines, or research funders.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers of care</li> <li>• Principal investigators</li> </ul> | <p>Not defined</p>                                                           | <p>No</p> | <p>Yes</p> | <p>No</p> | <p>Yes</p> |

|                |                                                                                                                                                                                                       |                                                                                                                                                                         |                                        |                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                                                                   |     |    |     |     |
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|                |                                                                                                                                                                                                       |                                                                                                                                                                         |                                        | <ul style="list-style-type: none"> <li>• Payers of research</li> </ul>                                                                                                                                                                                                                                                                            |                                                                                                                                                                                   |     |    |     |     |
| Dewidar 2022   | <p>Guidance informed by a series of methodological meetings, review of internationally renowned guidance (E.g. PRISMA), and review of exemplar rapid reviews on COVID-19</p> <p>Canada, 2020-2022</p> | To provide guidance for considering equity in rapid reviews through examples of published COVID-19 rapid reviews.                                                       | Rapid reviews                          | <p>A stakeholder is defined as an "individual or group responsible for or affected by health- and healthcare-related decision", including members of the public.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Public</li> </ul>                                                                                                    | 'Engagement' includes conducting, designing and interpreting the review. This includes providing feedback, prioritizing topics, suggesting gray literature, and providing advice. | Yes | No | No  | Yes |
| Eales 2017     | <p>Commentary</p> <p>Author expertise and examples from recently offered training opportunities</p> <p>2017</p>                                                                                       | To describe, based on the author's professional experiences, challenges and training opportunities to engage stakeholders in the conduct and use of evidence syntheses. | Systematic reviews and maps            | <p>"Stakeholders" include reviewer authors, researchers, end-users and decision-makers</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Public</li> <li>• Payers of research</li> <li>• Principal investigators</li> <li>• Peer review editors</li> <li>• Review commissioners</li> </ul> | "Engagement", which includes direct and indirect communication leading to two-way information flow among different stakeholder groups                                             | Yes | No | Yes | No  |
| Fadlallah 2020 | Systematic review                                                                                                                                                                                     | To systematically review the literature                                                                                                                                 | Systematic reviews, rapid reviews, gap | "Stakeholders" as defined by the 7Ps                                                                                                                                                                                                                                                                                                              | Referred to as "involvement" in the                                                                                                                                               | Yes | No | No  | Yes |

|               |                                                                                                                               |                                                                                                                                                                           |                                                             |                                                                                                                                                                                                                                                                                                                                                                                                                                      |                                                                                                     |     |     |     |     |
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|               | n = 29 included studies<br><br>Up to 2019                                                                                     | for proposed approaches and exercises conducted to prioritize topics or questions for systematic reviews and other types of evidence syntheses in any health-related area | maps, overviews of systematic reviews, and scoping reviews. | framework:<br><br>Groups: <ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients/Public</li> <li>• Purchasers</li> <li>• Payers</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Principal investigators</li> </ul>                                                                                                                                                                                      | priority setting process                                                                            |     |     |     |     |
| Feldman 2019  | Methodological review<br><br>n=120 included reviews<br><br>Systematic reviews published between January 2011 and October 2015 | To assess the extent and characteristics of stakeholder engagement in published systematic and rapid reviews and to specifically determine reporting characteristics.     | Systematic reviews and rapid reviews.                       | Stakeholders are anyone affected by an issue and/or anyone who can provide input on the topic. This includes decision makers, health professionals and their organizations are targeted, citizen or patients, other researchers and the media.<br><br>Groups: <ul style="list-style-type: none"> <li>• Providers of care</li> <li>• Patients</li> <li>• Public</li> <li>• Principal investigators</li> <li>• Policymakers</li> </ul> | The terms “involvement” and “engagement” are used interchangeably.                                  | No  | Yes | No  | No  |
| Garritty 2023 | Methodological guidance                                                                                                       | To discuss the importance of knowledge user involvement, highlight potential ways to                                                                                      | Rapid reviews                                               | ‘Knowledge user (KU)’ refers to an individual who is likely to use the knowledge generated through research to                                                                                                                                                                                                                                                                                                                       | A continuum of KU “involvement” from receiving information about a review to leading the initiation | Yes | No  | Yes | Yes |

|            |                                                                                                                                                                                                         |                                                                                                                                                                 |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                 |                                                                                                                                                                                                                                                                              |    |     |    |     |
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|            |                                                                                                                                                                                                         | engage users, and detail stages of knowledge user involvement in the rapid review process.                                                                      |                    | <p>make informed decisions about health policies, programmes or practices. It can more broadly imply an individual or group who may be responsible for or affected by health-related and healthcare-related decisions that these reviews can inform.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Patients</li> <li>• Public</li> <li>• Program managers</li> </ul> | and completion of a review.                                                                                                                                                                                                                                                  |    |     |    |     |
| Gavin 2023 | <p>Retrospective evaluation</p> <p>Online survey and evaluation workshops</p> <p>June 2022 – February 2023</p> <p>n=11 survey respondents (n=6 public, n=2 professional, n=3 researchers)</p> <p>UK</p> | To evaluate the experiences of, and quality of the involvement process from the perspectives of service users, professionals and early career researchers (ECRs | Systematic reviews | <p>Individuals were considered 'public' stakeholders if they were living with rheumatoid arthritis (RA), or a partner/friend caring for someone with RA. Involved healthcare professionals were registered occupational therapists or rheumatologists caring for those with RA.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers of care</li> </ul>                                    | Public and health professional involvement (PHPI) is an active and informative partnership, whereby research is conducted 'with' or 'by' the public [and professionals] (i.e. patients, service users, carers [healthcare providers]), rather than 'about' or 'done' to them | No | Yes | No | Yes |

|               |                                                                                                                                                          |                                                                                                                                                                                                                                                                                                                                                                |                           |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |                                                                                            |     |     |     |    |
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| Gerlach 2020  | Document review<br><br>7 Institute for Clinical and Economic Review (ICER) reviews published between 2014 and 2016<br><br>USA                            | To understand the frequency and characteristics of written stakeholder engagement with ICER draft evidence reports. Specifically, to describe the number, type, and source of comments submitted in response to ICER draft reports, and to examine the associations between healthcare stakeholder type, comment type and frequency, and the therapeutic area. | Evidence reviews for HTAs | Stakeholder types include pharmaceutical manufacturer, patient or patient group, provider or provider group, government (state or federal), health plan or pharmacy benefit manager (PBM), employer or employer coalition, or other (including consultant and other organizations not fitting a particular stakeholder type)<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients</li> <li>• Purchasers</li> <li>• Product makers</li> </ul> | Stakeholder comments received during public review of draft ICER evidence reports          | No  | No  | No  | No |
| Gierisch 2019 | Qualitative, conducted 10 interviews with evidence synthesis researchers and 2 focus groups with patient stakeholder informants.<br><br>August 2018, USA | To explore views, barriers, resources, and perceived values of engaging patient advisors in a national program of evidence synthesis research.                                                                                                                                                                                                                 | Systematic reviews        | Patients and caregivers                                                                                                                                                                                                                                                                                                                                                                                                                                                          | Referred to as “engagement”, “involvement”, and “collaborators” without further definition | No  | Yes | Yes | No |
| Gould 2017    | Case study of a systematic                                                                                                                               | To consider how knowledge translation                                                                                                                                                                                                                                                                                                                          | Systematic reviews        | "Stakeholders" include technical assistance                                                                                                                                                                                                                                                                                                                                                                                                                                      | Referred to as “engagement” or                                                             | Yes | Yes | No  | No |

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|               | review on disability policy research which involved policy stakeholders<br><br>USA                                     | strategies can support and advance systematic reviews that include diverse types of research.                                                                                                     |                                      | providers, policymakers, and academics. Other stakeholder categories that may benefit or use such research are people with disabilities, disability advocates, practitioners, and governmental agencies as well as scholars outside of the academy                      | "involvement" without further definition.                                            |     |     |     |     |
| Gribble 2014  | Systematic review<br><br>n= 107 included studies<br><br>Up to September 2012<br><br>USA                                | To describe the reporting of research ethics and community involvement in systematic reviews and meta-analyses concerning American Indian, Alaskan Native, and Native Hawaiian (AI/AN/NH) health. | Systematic reviews and meta-analyses | "Stakeholders" include AI/AN/NH tribes and communities, researchers, and journal editors<br><br>Groups:<br><ul style="list-style-type: none"> <li>Principal investigators</li> <li>Peer review editors</li> <li>Public</li> </ul>                                       | Referred to as "community involvement" and "consultation" without further definition | No  | No  | No  | Yes |
| Haddaway 2017 | Methodological framework based on a non-systematic literature review, key informant interviews, and author experience. | To provide a toolbox of possible stakeholder engagement activities, whilst also recommending approaches from stakeholder engagement research that may prove to be particularly useful for reviews | Systematic reviews                   | "Stakeholders": Any person or organization who can affect or may be affected by the planning, conduct, results and communication of a systematic review or map, including direct users of research outputs, such as policy decision makers and practitioners, and those | Referred to as "engagement" without further definition                               | Yes | Yes | Yes | Yes |

|               |                                                                                                                  |                                                                                                                                                                                                                                             |                   |                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                              |     |     |    |     |
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|               |                                                                                                                  |                                                                                                                                                                                                                                             |                   | directly affected by decisions (e.g. patients).                                                                                                                                                                                                                                                                                                        |                                                                                                                                              |     |     |    |     |
| Hannigan 2022 | Case study of a systematic review with stakeholder engagement<br><br>n=104 included studies<br><br>UK            | The aim of public and patient involvement [in this review] was to embed public and patient involvement perspectives into all aspects of the project, from the definition of search terms and parameters onwards to impact and dissemination | Systematic review | Stakeholders, including members of the public and people with personal experience of mental health and/or end of life care. Included professionals from a range of practitioner backgrounds based in NHS and charitable organizations, policy advisers and people with lived experience of mental health difficulties and of life-threatening disease. | Referred to as "public and patient involvement" described as "working with stakeholders" and included "equal partners" and "advisory groups" | No  | Yes | No | No  |
| Harris 2016   | Methodological guidance based on the author's experience of completing a review of community-based peer support. | To describe how methods used in participatory health research were used to involve patients, clients, providers and community health workers across all stages of a realist review.                                                         | Realist review    | Patient and public involvement (PPI) is an umbrella term, used to refer to a variety of stakeholders or end users/recipients/beneficiaries of research and means working with patients and the public as members of the research team. Article also uses the term "stakeholders" without further definition.<br><br>Groups:<br>• Patients              | Referred to as "engagement", "participation" and "involvement"                                                                               | Yes | No  | No | Yes |



|               |                                                                                                                                                                                                                          |                                                                                                                                                                  |                    |                                                                                                                                                                                                                                                                                                                                  |                                                                                                                                                                                                                                                                                                                                                                                                                                      |     |    |     |     |
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|               |                                                                                                                                                                                                                          |                                                                                                                                                                  |                    | <ul style="list-style-type: none"> <li>• Public</li> <li>• Policymakers</li> <li>• Providers</li> <li>• Principal investigators</li> <li>• Commissioners</li> </ul>                                                                                                                                                              |                                                                                                                                                                                                                                                                                                                                                                                                                                      |     |    |     |     |
| Harris 2018   | Methodological guidance based on author's practical experiences reviewing and developing methods for qualitative and mixed methods synthesis, and experiences conducting studies looking at the impact of participation. | To advance methods for systematically reviewing the impact of participation in health research, drawing on recent systematic review guidance.                    | Systematic reviews | People who are experienced in using participatory approaches in research and knowledgeable about conceptualisation of impact. This would include nonacademic social actors who are participating as coresearchers in guiding the participatory research projects. For example, people with lived experience of the health issue. | Defines "participation" as the extent to which a person or a group of people exert influence on health research, health structures, practices, services, or policies that have an effect on their health and well-being. It has been described alternatively as patient and public involvement and community engagement, with a range of influence possible, from minimal to being an equal partner in the research decision-making. | Yes | No | Yes | Yes |
| Harrison 2012 | Case study; Overview of a proposed academic-practice-policy partnership to address patient safety issues<br><br>Canada                                                                                                   | To describe the conceptual grounding of Queen's Joanna Briggs Collaboration for Patient Safety (the Partnership), its proposed methods, and its plan for action. | Systematic reviews | <p>"Stakeholders" referred to as policy or practice partners</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Principal investigators</li> </ul>                                                                                                                         | Integrated knowledge translation approach: engages researchers and research users throughout the research process, from research question to methodological choices, and involves them in the actual conduct of the reviews                                                                                                                                                                                                          | Yes | No | No  | No  |

|             |                                                                                                                                                                       |                                                                                                                                                                                                                                                       |                                                                               |                                                                                                                                                                                                                                                                                                                                 |                                                                                                                                                                                                                                                                                                              |    |     |    |    |
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|             |                                                                                                                                                                       |                                                                                                                                                                                                                                                       |                                                                               |                                                                                                                                                                                                                                                                                                                                 | or guideline adaptation project.                                                                                                                                                                                                                                                                             |    |     |    |    |
| Haynes 2010 | Case study of research methods for a decision-maker-researcher partnership systematic review<br><br>Canada                                                            | To form a partnership of healthcare providers, managers, and researchers to review randomized controlled trials assessing the effects of computerized decision support for six clinical application area.                                             | Systematic reviews                                                            | Decision-makers from the hospital and health region and the second is the research staff. This group is comprised of physicians, pharmacists, research staff, graduate students, and undergraduate students.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Providers</li> <li>• Principal investigators</li> </ul> | The role of the decision-makers is to guide the review process. The role of the research staff is to do the work 'in the trenches,' that is, undertake a comprehensive literature search, extract the data, synthesize the data, plan dissemination, and engage in the partnership.                          | No | Yes | No | No |
| Heenan 2023 | Two case studies: the first drawing on a body of work in public health law, and the second on a body of work focused on the first 2000 days of life.<br><br>Australia | To describe the development of a knowledge synthesis approach that utilizes coproduction with policymakers to combine the findings of a programme of research with policy knowledge to support decision-makers working in chronic disease prevention. | Knowledge synthesis processes, including systematic reviews and meta-analyses | Specific to policymakers through the use of policy dialogues.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Policymakers</li> </ul>                                                                                                                                                                                | Coproduction: a mechanism where stakeholders collaborate, generate knowledge that is relevant to the context and apply it in practice. Different variations of coproduction (for example, codesign, cocreate, coproduce) are used; however, "co" is suggestive of the collaborative or participatory design. | No | Yes | No | No |
| Hempel 2019 | Case study of                                                                                                                                                         | To describe a                                                                                                                                                                                                                                         | Evidence reviews                                                              | Referred to as                                                                                                                                                                                                                                                                                                                  | Referred to as "input"                                                                                                                                                                                                                                                                                       | No | Yes | No | No |

|              |                                                                                                                                                                                       |                                                                                                                                                                                                                                                                                                                                                                                                                                             |                                                                                                                              |                                                                                                                                                                                                                                                                                            |                                                                                                  |     |     |     |    |
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|              | <p>scoping searches, stakeholder input, and feasibility scans. The project is ongoing; this manuscript describes methods and results from June 2016 to September 2018.</p> <p>USA</p> | <p>structured and transparent approach to identify and prioritize areas of psychological health that are important and that can be feasibly addressed by a synthesis of the research literature. It describes a collaboration between an agency charged with facilitating the implementation of evidence-based research and practices across the Military Health System (MHS) and a research center specializing in evidence synthesis.</p> |                                                                                                                              | <p>"stakeholders" without further definition</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Principal investigators</li> </ul>                                                                                                                           | <p>whereby stakeholders could rate topic priorities and suggest topics for evidence reviews.</p> |     |     |     |    |
| Hoffman 2010 | <p>Qualitative assessment of case studies through documents and semistructured interviews</p> <p>Baltimore, USA</p>                                                                   | <p>To describe and assess our experiences with engaging stakeholders in comparative effectiveness research, to provide useful insights for others doing similar work.</p>                                                                                                                                                                                                                                                                   | <p>Comparative effectiveness research, including both the synthesis of existing evidence and the production of new data.</p> | <p>"Stakeholders" responsible for individual health care choices, professional policies, and reimbursement decisions.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients</li> <li>• Payers</li> <li>• Purchasers</li> <li>• Policymakers</li> </ul> | <p>Referred to as "engagement" without further definition</p>                                    | Yes | Yes | Yes | No |

|              |                                                                                                                                            |                                                                                                                                                                                                                                                                                                                       |                    |                                                                                                                                                                                                              |                                                                                                                                                                                                                                       |    |     |     |    |
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| Hyde 2017    | Case study of a systematic review and researcher reflections on how patient and public involvement modified the review at each time-point. | To describe the process and impact of involving patients in a systematic review and narrative synthesis of shared decision making around prescribing analgesia for musculoskeletal pain in primary care consultations.                                                                                                | Systematic reviews | Patients and the public, described as “service users”<br>Groups: <ul style="list-style-type: none"><li>• Public</li><li>• Patients</li></ul>                                                                 | “Involvement” has been defined as ‘doing research ‘with’ or ‘by’ the public, rather than ‘to’, ‘about’ or ‘for’ the public’, and patient engagement as ‘where information and knowledge about research is provided and disseminated’. | No | Yes | Yes | No |
| Jacaban 2022 | Cross-sectional analysis of existing reviews and online qualitative survey of review authors.<br><br>2019-2020                             | To describe the landscape of environmental systematic reviews and describe SR authors views on applications of their work, including possible barriers and facilitators for application, as well as how authors of SRs engage with stakeholders and communicate findings to maximize the impact of environmental SRs. | Systematic reviews | Referred to as “stakeholders” and “end-users” including decision-makers, policy-makers, and practitioners.<br><br>Groups: <ul style="list-style-type: none"><li>• Providers</li><li>• Policymakers</li></ul> | Referred to as "engagement" without further definition                                                                                                                                                                                | No | Yes | Yes | No |
| Janke 2022   | Scoping review with community partners<br><br>n=141 included studies                                                                       | To curate and describe the partnership literature in the community engagement field in order to advance partnership research                                                                                                                                                                                          | Scoping review     | Community partners (formal and informal groups) including: non-profit organizations, government, industry/business, faith-based organizations,                                                               | The collaboration between institutions of higher education and their larger communities (local, regional/state, national, global) for the mutually                                                                                    | No | Yes | No  | No |

|             |                                                                                       |                                                                                                                                                                                                                                                                                                                                                                                                                         |                    |                                                                                                                                                                                                  |                                                                                            |     |     |     |    |
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|             |                                                                                       | and practice. The guiding question was "What is the state of the partnership literature in the field of community engagement?"                                                                                                                                                                                                                                                                                          |                    | health, or international partners.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Program managers</li> </ul> | beneficial exchange of knowledge and resources in a context of partnership and reciprocity |     |     |     |    |
| Kelson 1999 | Cross-sectional questionnaire of Cochrane Review Groups<br><br>January 1998<br><br>UK | To identify the extent to which the Cochrane Collaboration involves consumers (patients, carers, members of patient/consumer organizations) as members of Cochrane Review Groups (CRGs); to explore the emphasis CRGs place on identifying and collecting information on outcomes identified by patients as being important indicators of quality and effectiveness of treatment and care ("patient-defined outcomes"). | Systematic reviews | Consumers (patients, carers, and members of patient organizations)<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> </ul>                                                | Referred to as "involvement" and "participation"                                           | No  | No  | No  | No |
| Keown 2008  | Description of stakeholder engagement activities based                                | To describe and discuss various stakeholder engagement                                                                                                                                                                                                                                                                                                                                                                  | Systematic reviews | A consumer of the research findings, an audience member who has a stake in the topic                                                                                                             | Referred to as "engagement" without further definition.                                    | Yes | Yes | Yes | No |

|             |                                                                                                           |                                                                                                                                                                                   |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                        |                                       |     |     |     |     |
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|             | on 22 systematic reviews involving more than 80 stakeholders over a 4-year period<br><br>Canada           | opportunities that we employ throughout the stages of conducting a systematic review, to increase knowledge utilization within interested non research audiences or stakeholders. |                    | being investigated, or a potential decision maker (policy and/or practice) within the context of their role. Specific stakeholder groups include policy and practitioner decision makers from worker compensation boards, government, health and safety consultancy, clinical settings, organized labor, and employer and injured worker groups.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Program managers</li> <li>• Public</li> </ul> |                                       |     |     |     |     |
| Kreis 2013  | In-depth, semi-structured interviews with key informants and review of organizations websites.<br><br>USA | To explore current approaches to involving consumers in US-based and key international organizations and groups conducting or commissioning systematic reviews (organizations).   | Systematic review  | “Consumers” include patients, families of patients, lay caregivers, consumer advocates and members of the general public<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Public</li> <li>• Patients</li> </ul>                                                                                                                                                                                                                                                                              | Referred to as “consumer involvement” | Yes | Yes | Yes | No  |
| Langer 2017 | Commentary                                                                                                | To address head on                                                                                                                                                                | Systematic reviews | Stakeholders not                                                                                                                                                                                                                                                                                                                                                                                                                                                                                       | A range of approaches                 | Yes | No  | No  | Yes |

|           |                                                                                                                                                            |                                                                                                                                                                                                                                                           |                                                         |                                                                                                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                                                                    |     |     |     |    |
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|           | Author experiences from the Africa Centre for Evidence at the University of Johannesburg South Africa                                                      | the often undiscussed key challenge with regard to stakeholder involvement in systematic reviews: that responding to stakeholders can mean reconsidering what makes a review rigorous.                                                                    |                                                         | further defined.                                                                                                                                                                                                                                                                                                                                                                                    | for “engagement” in research, from advisory groups to co-production. This range includes the opportunity for stakeholders to shape the scope of the review, the types of outcomes considered, and the dissemination of the research findings, amongst other things |     |     |     |    |
| Land 2017 | Methodological description of the five-step approach to stakeholder engagement in the review prioritization and planning stage.<br><br>2012-2017<br>Sweden | To report on the empirically tested five-step approach that the Mistra Council for Evidence-based Environmental Management (EviEM) is using to engage stakeholders and incorporate their views and opinions in the prioritization and planning of reviews | Systematic reviews and systematic maps                  | “Stakeholders” are those who use or may be affected by review findings, including researchers and subject experts, practitioners, commissioners, and representatives of governmental and non-governmental organizations<br><br>Groups: <ul style="list-style-type: none"> <li>• Providers</li> <li>• Principal investigators</li> <li>• Review commissioners</li> <li>• Program managers</li> </ul> | “Engagement” is defined as a bi-directional relationship between the stakeholder and the researcher that results in informed decision-making about the selection, conduct, and use of research findings.                                                           | Yes | No  | Yes | No |
| Lips 2022 | Document analysis of all drug                                                                                                                              | To analyze stakeholder involvement in                                                                                                                                                                                                                     | Relative effectiveness assessments of health technology | “Stakeholders” without further definition. Specifically highlights                                                                                                                                                                                                                                                                                                                                  | “Involvement” without further definition.                                                                                                                                                                                                                          | Yes | Yes | Yes | No |

|             |                                                                                                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                  |                                                                                                                                      |                                                                                                                                             |                                                                                                                                                                                                                                                                                                                                                  |     |     |     |     |
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|             | <p>assessments completed in 2019 by the public HTA agency of The Netherlands, enriched with semistructured interviews with employees of the HTA agency and representatives of patient and medical associations involved in these assessments.</p> <p>Netherlands 2019</p> | <p>assessing relative effectiveness and how the parties involved value this collaboration.</p> <p>Research questions:</p> <p>1. What role do patients and medical professionals have in the realization of a relative effectiveness assessment (REA)?</p> <p>2. How is this cooperation experienced and valued by the involved stakeholders?</p> | assessments (HTAs).                                                                                                                  | <p>patients and medical professionals.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients</li> </ul> |                                                                                                                                                                                                                                                                                                                                                  |     |     |     |     |
| Manafo 2018 | <p>Rapid systematic review</p> <p>n= 70 articles</p> <p>2007-2017</p> <p>Canada, the US, Europe, UK, Australia/New Zealand, or Scandinavian countries</p>                                                                                                                 | <p>To describe the evidence that does exist in relation to patient and public engagement priority setting in both health ecosystem and health research</p>                                                                                                                                                                                       | <p>'Pre-preparation' and 'preparation' phases of health ecosystem and research priority setting (e.g., Global Evidence Mapping).</p> | Patients and the public                                                                                                                     | <p>"Engagement" focusses on what knowledge is (or questions are) valued most by patients and the public as they become experts in their health care experiences. The authors identify two levels of engagement:</p> <p><i>Deliberative:</i> Public and Patients collaborate by co- developing topics for research with researchers and other</p> | Yes | Yes | Yes | Yes |



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|             |                                                                   |                                                                                                                                                                                                                                                            |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                   | <p>key groups</p> <p><i>Consultative:</i> Public and Patients consult about research topics and priorities that are most important to them.</p>                                                             |     |     |     |    |
| Mann 2019   | <p>Description of rapid review methodology</p> <p>Wales (UK)</p>  | To describe the rapid systematic review approach used by the Palliative Care Evidence Review Service (PaCERS)                                                                                                                                              | Rapid reviews      | <p>Referred to as “stakeholders”, who are described as clinicians, service users and policymakers.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients</li> <li>• Policymakers</li> <li>• Public</li> </ul>                                                                                                                                                                                 | Referred to as “engagement” and “working in partnership”.                                                                                                                                                   | Yes | No  | No  | No |
| Merner 2021 | <p>Series of case studies (n=4)</p> <p>Cochrane Collaboration</p> | To share the stakeholder involvement experiences of three Cochrane editorial groups (Consumers and Communication, Effective Practice and Organization of Care, and Public Health) that form part of the Cochrane Public Health and Health Systems Network. | Systematic reviews | <p>“Stakeholders” defined as policymakers, health decision-makers, health professionals, consumers (meaning patients, caregivers, their representatives, and the public), guideline developers, and research funders</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Policymakers</li> <li>• Patients</li> <li>• Public</li> <li>• Review commissioners</li> <li>• Payers of research</li> </ul> | “Involvement” is any role or contribution of stakeholders toward the development of a review protocol, completion of any of the stages of a systematic review, or dissemination of the findings of a review | No  | Yes | Yes | No |

|             |                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                                             |                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                |    |     |     |     |
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| Mogo 2020   | Case study of a rapid review with stakeholder engagement to define priority areas.<br><br>n=26 included studies<br><br>2017<br><br>Canada | To contribute to the existing knowledge on how best to mobilize research into policy by detailing a process which engaged in an iterative and collaborative process of generating and adapting research to inform decision-making, involving the input of grassroots organizations, multi-sectoral stakeholders working on issues of childhood disability and policymakers. | Rapid reviews      | "Stakeholders" under the umbrella of CHILD LeisureNet – a network of various members of the childhood disability community that range from health and education professionals, parents of children with disabilities, youth with disabilities, policymakers and community grassroots organization<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Providers</li> <li>• Patients</li> <li>• Program managers</li> </ul> | Referred to as "engagement" without further definition. Authors also describe their approach as "integrated knowledge translation"                                                                                                                                                             | No | Yes | No  | Yes |
| Morley 2016 | Scoping review<br><br>n=36 included studies<br><br>1990-2015                                                                              | To systematically scope the evidence base for consumer involvement in organizations which commission, undertake or support systematic reviews; with an emphasis on Cochrane.                                                                                                                                                                                                | Systematic reviews | "Healthcare consumers" who use Cochrane evidence are generally patients, carers and family members, or people interested in remaining healthy who are seeking information about a health condition or treatment for personal use.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> </ul>                                                                                                                                          | Referred to as "involvement". The types of involvement which were documented could be broken down into four main categories: formal roles in review production and coordination, roles in review production, roles in increasing the accessibility of review, and roles in knowledge transfer. | No | Yes | Yes | Yes |

|                  |                                                                                          |                                                                                                                                                                                                                                       |                    |                                                                                                                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                                                             |     |    |    |     |
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|                  |                                                                                          |                                                                                                                                                                                                                                       |                    | <ul style="list-style-type: none"> <li>Public</li> </ul>                                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                                             |     |    |    |     |
| Morley 2022      | Gray literature guidance document                                                        | To set out issues for Cochrane review authors on how to involve healthcare consumers (patients, carers, and the public) in funding applications.                                                                                      | Systematic reviews | <p>"Consumers" can be patients, carers, family members or the public.</p> <p>Health professionals or people using systematic reviews to inform health policy or practice may also be involved. This may be called stakeholder involvement.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>Patients</li> <li>Public</li> <li>Providers of care</li> <li>Policymakers</li> </ul>    | "Involvement" (sometimes called patient and public involvement, engagement, co-production and other terms) is the meaningful involvement of people affected by a healthcare condition in research or the delivery of activities or services | Yes | No | No | No  |
| Munthe-Kaas 2022 | Qualitative user-tests (n=4) of the TRANSFER conversation guide<br><br>n=17 participants | To explore the user experience of the TRANSFER approach conversation guide, and in doing so gain a better understanding of the role and perceived value of stakeholder engagement in systematic reviews for informed decision-making. | Systematic reviews | <p>Stakeholders refers to anyone with an interest in the findings from a systematic review, including patients and service users, patients and the public, health care practitioners, policymakers and other decision makers in the fields of health, education, social welfare, and environmental protection.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>Patients</li> </ul> | Referred to as "engagement" and "collaboration" without further definition                                                                                                                                                                  | Yes | No | No | Yes |

|               |                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                   |                                                 |                                                                                                                                                                                             |                                                                                                                            |     |     |    |     |
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|               |                                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                   |                                                 | <ul style="list-style-type: none"> <li>• Public</li> <li>• Providers of care</li> <li>• Policymakers</li> </ul>                                                                             |                                                                                                                            |     |     |    |     |
| Nasser 2013   | <p>Methodology paper; Development of the equity lens for priority setting framework using: (1) a workshop presenting survey results from a previous project, (2) a review of the literature, and (3) a final workshop for refinement of the equity lens</p> <p>2008 &amp; 2009 Cochrane Colloquium (conference)</p> | To develop and pilot an equity lens that could help researchers in developing a more equity-oriented approach toward priority setting and agenda setting in systematic reviews.                   | Priority setting for systematic reviews         | <p>"Stakeholders" without further definition</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers</li> <li>• Policymakers</li> </ul> | Referred to as "engagement" without further definition.                                                                    | Yes | No  | No | Yes |
| O'Connor 2023 | <p>Case study of a priority setting exercise using surveys and workshops</p> <p>n=68 survey respondents from 10 countries</p>                                                                                                                                                                                       | 1) To rapidly identify priority topic uncertainties as a first step to identify future systematic review questions of pertinence to key international fecal incontinence (FI) stakeholders. 2) To | Priority setting for Cochrane systematic review | <p>Referred to as "stakeholders" including patients, carers, health care professionals, policy makers and voluntary, community, or social enterprise representatives.</p> <p>Groups:</p>    | <p>Patient and public involvement. Involvement was at an "influencing" level as described within the ACTIVE Framework.</p> | No  | Yes | No | Yes |

|                |                                                                                                                                                                     |                                                                                                                                                                                                                                          |                                                                                                                                                                                  |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                           |                                                                                                                 |     |    |     |    |
|----------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------|-----|----|-----|----|
|                |                                                                                                                                                                     | share our methods, experience, and learning with other groups planning to deliver a rapid priority setting exercise.                                                                                                                     |                                                                                                                                                                                  | <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers of care</li> <li>• Policymakers</li> <li>• Public</li> </ul>                                                                                                                                                                                                                                                                                                                                                       |                                                                                                                 |     |    |     |    |
| Parker 2020    | <p>Qualitative; Gray literature report</p> <p>Interviews (2020-2021) with members of each of the eight Cochrane Review Groups which constitute the MOSS network</p> | 1) To understand the current stakeholder engagement work within MOSS, 2) to share examples of stakeholder engagement work within MOSS, and 3) to gather information to inform the next steps in stakeholder engagement work within MOSS. | Systematic reviews                                                                                                                                                               | <p>A “stakeholder” is an individual or group who is responsible for, or affected by health- and healthcare-related decisions. Examples of stakeholders included authors, consumers, journals, academic institutions, policy makers, and guideline developers.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Peer review editors</li> <li>• Principal investigators</li> <li>• Policymakers</li> <li>• Review commissioners</li> </ul> | “Engagement” is defined as an “active partnership” between stakeholders and researchers in the research process | No  | No | Yes | No |
| Petticrew 2023 | Methodological guidance                                                                                                                                             | To present the Commercial Determinants of Health and Evidence Synthesis (CODES) methodological guidance for                                                                                                                              | Evidence synthesis, which includes a wide range of types of review including both narrative systematic reviews and meta-analyses, as well as scoping, mapping and other reviews. | <p>Referred to as “stakeholders”, with a focus on industry stakeholders and patients/public.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> </ul>                                                                                                                                                                                                                                                                                                              | Referred to as “co-production”, “partnership” and “patient/public involvement”                                  | Yes | No | No  | No |

|              |                                                                                                                                                                                                       |                                                                                                                                                                                                                                    |                    |                                                                                                                                                                                                                                                                                                 |                                                                                                                                                                                                                                     |     |     |    |    |
|--------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|-----|----|----|
|              |                                                                                                                                                                                                       | systematic reviews and other evidence syntheses                                                                                                                                                                                    |                    | <ul style="list-style-type: none"> <li>Public</li> <li>Product makers</li> </ul>                                                                                                                                                                                                                |                                                                                                                                                                                                                                     |     |     |    |    |
| Pollock 2015 | Case study;<br>Three meetings during the course of updating a Cochrane systematic review. Used nominal group techniques to reach consensus on the structure and methods of the review<br><br>Scotland | To describe the structured methods used to involve patients, carers and health professionals in an update of a Cochrane systematic review relating to physiotherapy after stroke and explores the perceived impact of involvement. | Systematic reviews | <p>Refers to "stakeholders" without further definition. Also identifies 'consumer' as a person with a healthcare condition, or friend, carer (unpaid) or family member of that person.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>Patients</li> <li>Providers</li> </ul>         | "Involvement" without further definition                                                                                                                                                                                            | No  | Yes | No | No |
| Pollock 2018 | Scoping review<br><br>n=291 included studies<br><br>2010-2016                                                                                                                                         | To document the evidence-base relating to stakeholder involvement in systematic reviews; and use this evidence to describe key features of how stakeholders have been involved in systematic reviews                               | Systematic reviews | <p>A "stakeholder" is any person who would be a knowledge user of research but whose primary role is not directly in research.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>Patients</li> <li>Public</li> <li>Providers</li> <li>Policymakers</li> <li>Program managers</li> </ul> | "Involvement" in a systematic review is any role or contribution of stakeholders toward the development of a review protocol, completion of any of the stages of a systematic review, or dissemination of the findings of a review. | Yes | Yes | No | No |
| Pollock 2019 | Scoping review and workshop                                                                                                                                                                           | To report the development of the                                                                                                                                                                                                   | Systematic reviews | A "stakeholder" is any person who uses                                                                                                                                                                                                                                                          | "Involvement" in a systematic review is                                                                                                                                                                                             | Yes | No  | No | No |

|              |                                                                                                                                                                                |                                                                                                                                                                         |                                                          |                                                                                                                                                                                                                                                                                                                      |                                                                                                                                                                                             |     |    |     |     |
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|              | UK                                                                                                                                                                             | ACTIVE framework to describe how stakeholders are involved in systematic reviews                                                                                        |                                                          | research knowledge but whose primary role is not directly in research.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers</li> <li>• Policymakers</li> <li>• Program managers</li> </ul>                                                                     | any role or contribution of stakeholders toward the development of a review protocol, completion of any of the stages of a systematic review, or dissemination of the findings of a review. |     |    |     |     |
| Pollock 2022 | Guidance paper developed by the JBI Scoping Review Methodology Group.<br><br>Based on the available evidence, and the experiences of the JBI Scoping Review Methodology Group. | To present JBI's guidance for knowledge user engagement in scoping reviews based on the expert opinion of the JBI Scoping Review Methodology Group.                     | Scoping reviews                                          | "Knowledge users" are those invested in the production of research, and who may benefit or be impacted by the research.<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Patients</li> <li>• Principal investigators</li> <li>• Providers</li> <li>• Policymakers</li> <li>• Payers of research</li> </ul> | "Engagement" is a bi-directional relationship between stakeholder and researcher that results in informed decision-making about the prioritization, conduct and use of research.            | Yes | No | Yes | Yes |
| Premji 2022  | Cross-sectional online survey to four email lists with broad participation among business librarians across the globe                                                          | To assess the following areas:<br>1) the current knowledge synthesis awareness among business librarians; 2) their involvement in various types of knowledge synthesis; | Systematic reviews and other knowledge synthesis reviews | Academic business librarians<br><br>Groups:<br><ul style="list-style-type: none"> <li>• Principal investigators (and members of the research team)</li> </ul>                                                                                                                                                        | "Involvement" without further definition                                                                                                                                                    | Yes | No | Yes | No  |

|             |                                                                                                                                                                                                           |                                                                                                                                                                                        |                                                                                                |                                                                                                                                                                                                                                                                                                                                                                                                                                       |                                                                                                                                                                                                                                                     |     |     |     |    |
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|             | n=71<br>respondents<br><br>2020                                                                                                                                                                           | 3) the level of training and training formats used to develop competencies; and 4) barriers to participation among librarians supporting academic programs of business and management. |                                                                                                |                                                                                                                                                                                                                                                                                                                                                                                                                                       |                                                                                                                                                                                                                                                     |     |     |     |    |
| Saan 2015   | Employs the Tool for Recording and Accounting for Stakeholder Involvement (TRASI) in two cases of systematic reviews (quantitative and qualitative) that are presented as worked examples (case studies). | The aim of this study was to describe some of the processes of involvement of stakeholders and to demonstrate a Tool for Recording and Accounting for Stakeholder Involvement (TRASI)  | Systematic reviews (referring to both quantitative reviews and qualitative evidence syntheses) | <p>“Stakeholders” have some self-interest in the review, either because they are going to use the findings of the review or because decisions made by others in light of the findings might have an impact on them.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Providers</li> <li>• Patients</li> <li>• Public</li> <li>• Review commissioners</li> <li>• Principal investigators</li> </ul> | Four approaches describe stakeholder involvement in systematic reviews: Stakeholders taking control, stakeholders collaborating with researchers, stakeholders being consulted, and stakeholders having a minimal interaction with the researchers. | Yes | Yes | No  | No |
| Sakala 2001 | Case study of Cochrane’s Pregnancy and Childbirth Group                                                                                                                                                   | To describe an innovative pilot project to involve consumers and consumer advocates in the process of refereeing systematic                                                            | Systematic reviews                                                                             | <p>Referred to as “consumers”</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> </ul>                                                                                                                                                                                                                                                                                                       | “Involvement” without further definition                                                                                                                                                                                                            | No  | No  | Yes | No |



|                |                                                                                                                                                                          |                                                                                                                                                                |                                         |                                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                                                                                                                                                                   |    |     |     |    |
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|                |                                                                                                                                                                          | reviews in preparation by members of the Cochrane Collaboration's Pregnancy and Childbirth Group.                                                              |                                         |                                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                                                                                                                                                                   |    |     |     |    |
| Sanderson 2022 | Case study; (1) a description of the iKT process; (2) the results of the iKT engagement as they map onto these steps; and (3) a methodological discussion.<br><br>Canada | To describe how an integrated knowledge translation (iKT) approach was embedded within a master's thesis project comprising a mixed-methods systematic review. | Mixed-method systematic review          | 'Knowledge user' describes individuals who are likely to use research results to make informed decisions, but similar terms are also used (eg, stakeholders, decision-makers).<br><br>Groups: <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Program Managers</li> <li>• Providers</li> <li>• Patients</li> <li>• Principal Investigators</li> </ul> | iKT is a model of collaborative research, where researchers work with knowledge users (KUs), who identify a problem and have the authority to implement the research recommendations<br>iKT involves KUs – as equal partners – who play an integral role in the research process. | No | Yes | Yes | No |
| Scott 2018     | Two-phase international online survey<br><br>2017-2018                                                                                                                   | To report the results of a prioritization project, aiming to identify highest priority systematic review topics.                                               | Priority setting for systematic reviews | "Stakeholders" were survey respondents. No definition is provided.<br><br>Groups: <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Providers</li> <li>• Patients</li> <li>• Payers of research</li> <li>• Principal investigators</li> <li>• Review commissioners</li> </ul>                                                                           | Referred to as "involvement" without further definition                                                                                                                                                                                                                           | No | Yes | No  | No |

|                   |                                                                                                                                                                                              |                                                                                                                                                                                                                                                     |                             |                                                                                                                                                                                                                                         |                                                                                                                                                                                                                                                                                                                                                     |     |     |     |     |
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| Smith 2008 & 2009 | Guidance; Informed by author experiences of recruiting service users to a reference group for a review of user involvement in nursing, midwifery and health visiting research<br><br>England | To support the critical development of user involvement in systematic reviews by explaining some of the theoretical, ethical and practical issues entailed in getting ready for user involvement.                                                   | Systematic reviews          | Service users (patients, carers and the public)<br><br>Groups: <ul style="list-style-type: none"><li>• Patients</li><li>• Public</li></ul>                                                                                              | "Involvement" has hierarchical levels of control from consultation, through to collaboration and user-control. In this particular review, user involvement focused on interpretation and implementation of the findings to form guidance. Authors frame user involvement as 'relationships within social contexts'.                                 | No  | No  | Yes | Yes |
| Soobiah 2019      | Cross-sectional survey of engagement using the Patient Engagement Evaluation tool (PEET).<br><br>n=15 respondents<br><br>Canada                                                              | To quantify the perceived level of engagement experienced by knowledge users involved in different activities in the conduct of a systematic review on the comparative effectiveness of geriatrician-led models of care across health care settings | Systematic reviews          | Knowledge users (KUs) are individuals who may use research findings to make a decision.<br><br>Groups: <ul style="list-style-type: none"><li>• Patients</li><li>• Providers</li><li>• Policymakers</li><li>• Program managers</li></ul> | Integrated knowledge translation (iKT) is an approach whereby researchers and knowledge users co-create knowledge. Article also refers to "KU engagement" defined as including individuals who may be affected by the research findings through a process of meaningful involvement in making decisions about key aspects of the systematic review. | No  | Yes | No  | No  |
| Spök 2018         | Commentary<br><br>EU                                                                                                                                                                         | To describe the stakeholder involvement strategy developed and                                                                                                                                                                                      | Systematic reviews and maps | "Stakeholders" included competent authorities, industry, civil society organizations,                                                                                                                                                   | "Engagement"                                                                                                                                                                                                                                                                                                                                        | Yes | No  | Yes | No  |

|              |                   |                                                                                                                                                                                                                                                                                                 |                                                           |                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                 |     |     |    |    |
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|              |                   | experiences gathered during the development and prioritization of review questions of the GRACE project                                                                                                                                                                                         |                                                           | <p>professional organizations and researchers.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Product makers</li> <li>• Principal investigators</li> </ul>                                                                                              |                                                                                                                                                                                                 |     |     |    |    |
| Springs 2019 | Case study<br>USA | To outline and elaborate the approach for this project, innovating on well-established methods of evidence synthesis by: (1) training and mentoring community research partners; and (2) developing an online, interactive evidence map to support dissemination and implementation of evidence | Evidence synthesis methods, particularly evidence mapping | <p>“Stakeholders” as defined by the 7P’s framework.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Providers</li> <li>• Patients/Public</li> <li>• Purchasers</li> <li>• Payers</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Principal investigators</li> </ul> | “Community engaged approach” to evidence synthesis to codesign, coproduce and co-disseminate an evidence map.                                                                                   | No  | Yes | No | No |
| Sundin 2018  | Commentary        | To present a framework for the integration of storytelling in systematic reviews and systematic maps at the stages where stakeholders are actively involved.                                                                                                                                    | Systematic reviews and systematic maps                    | <p>Stakeholders are defined as all individuals and organizations that might have a stake in the findings of the review.</p> <p>Groups not specified.</p>                                                                                                                                     | “Engagement” or “involvement” of stakeholders as either consultants or collaborators to the review team. As collaborators, the stakeholders are engaged to a larger extent than as consultants. | Yes | No  | No | No |

|                      |                                                                                                                      |                                                                                                                                                                                                                                                                                           |                    |                                                                                                                                                                                                                                                                                                                                                                                                            |                                                                                                                                                                                                                                                                   |     |     |     |     |
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| Synnot 2019 & 2020   | <p>Priority setting workshop and a post-hoc evaluation of the process.</p> <p>n=28 participants</p> <p>Australia</p> | <p>To select and refine priority topics for systematic reviews in health communication and participation, and use these to identify five priority Cochrane Consumers and Communication Group (CCCG) Reviews</p> <p>To describe the evaluation of the project's processes and outcomes</p> | Systematic reviews | <p>Consumers and other stakeholders</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers</li> <li>• Policymakers</li> <li>• Payers of health services</li> <li>• Payers of research</li> <li>• Principal investigators</li> <li>• Program managers</li> </ul>                                                                                       | Working in partnership, informed by the principles of co-production (sharing of power and responsibility, including all perspectives and skills, respecting and valuing contributors' knowledge, and cultivating mutually beneficial and supportive relationship) | No  | Yes | No  | Yes |
| Taylor 2017          | <p>Author reflections based on experiences with government commissioned reviews</p> <p>Canada</p>                    | To provide five lessons to consider when undertaking a systematic review with stakeholders that are new to the process to help ensure a successful relationship between the review team and the stakeholders involved.                                                                    | Systematic reviews | <p>"Stakeholders" include all parties that may affect or be affected by a review. For the purpose of this commentary, author advice applies mainly to those involved in the review process from question definition to review synthesis (i.e., commissioners, funders, advisors)</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Review commissioners</li> <li>• Payers of research</li> </ul> | "Engagement" without further definition                                                                                                                                                                                                                           | No  | No  | Yes | No  |
| Todhunter-Brown 2022 | Gray literature guidance                                                                                             | To1) highlight the importance of                                                                                                                                                                                                                                                          | Systematic review  | "Stakeholders" defined according to the 7Ps                                                                                                                                                                                                                                                                                                                                                                | "Involvement" includes two approaches:                                                                                                                                                                                                                            | Yes | No  | No  | No  |

|                |                                   |                                                                                                                                                                                                                                                                                                                                                                                                                                                          |                    |                                                                                                                                                                                                                                                                                                                                                 |                                                                                                                                                                                                                                                                                                            |     |    |    |    |
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|                | document                          | planning patient and public involvement (PPI) in a systematic review 2) describe who you might involve in your review 3) describe the stages when you might involve people 4) describe the different levels of involvement you might have 5) describe how people can be involved in a systematic review 6) provide a framework for describing and reporting how you involved people 7) signpost readers to a range of resources for further information. |                    | <p>framework.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers of care</li> <li>• Purchasers</li> <li>• Payers of health research</li> <li>• Policymakers</li> <li>• Principal investigators</li> <li>• Product makers</li> </ul>                                                    | <p>Continuous involvement – people are involved ‘throughout’ the review process, perhaps as a member of the review author team or an advisory group.</p> <p>One-time involvement – people are involved at a specific stage in a review in order to complete a specific task or address a specific aim.</p> |     |    |    |    |
| Tomlinson 2021 | Gray literature guidance document | To present Cochrane’s six-step stakeholder engagement framework.                                                                                                                                                                                                                                                                                                                                                                                         | Systematic reviews | <p>Stakeholder is an individual or group who is responsible for, or affected by health- and healthcare-related decisions.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers of care</li> <li>• Program managers</li> <li>• Policymakers</li> <li>• Principal investigators</li> </ul> | Engagement is “an active partnership between stakeholders and researchers in the research process                                                                                                                                                                                                          | Yes | No | No | No |

|                |                                                                                                              |                                                                                                                                                                                                      |                                                                      |                                                                                                                                                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                                                                                                                |     |     |     |    |
|----------------|--------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|-----|-----|----|
|                |                                                                                                              |                                                                                                                                                                                                      |                                                                      | <ul style="list-style-type: none"> <li>• Payers of health research</li> </ul>                                                                                                                                                                                                                                                                                                                                                |                                                                                                                                                                                                                                                                                                |     |     |     |    |
| Tomlinson 2023 | Case study of a priority setting exercise using the SPARK tool for priority setting<br><br>n=13 stakeholders | To pilot a priority setting exercise based on systematic reviews of interventions published in the Cochrane Library, to identify and prioritize reviews to be updated with a focus on health equity. | Cochrane systematic reviews                                          | <p>“Stakeholders” defined as an individual or group who is responsible for or affected by health- and healthcare-related decisions that can be informed by research evidence. This project involved one stakeholder representing patients/public, and the remainder were ‘researchers’.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Principal investigators</li> <li>• Patients</li> <li>• Public</li> </ul> | Described as “engaged” and “working with”.                                                                                                                                                                                                                                                     | No  | Yes | No  | No |
| Tricco 2018    | Scoping review<br><br>n= 91 included studies<br><br>1996-2016                                                | To map the evidence on engaging knowledge users with an emphasis on policy-makers, health system managers, and policy analysts in the knowledge synthesis process                                    | Knowledge synthesis, such as a systematic review or a scoping review | “Knowledge users” are policymakers, policy analysts, and health system managers, at some level of government or decision-making institution, who have responsibility for making recommendations to others, analyzing data and informing decisions and recommendations,                                                                                                                                                       | Engagement, defined as an iterative process of actively soliciting the knowledge, experience, judgment and values of individuals selected to represent a broad range of direct interests in a particular issue, for the dual purposes of: creating a shared understanding and making relevant, | Yes | Yes | Yes | No |

|             |                                                                                                                                                           |                                                                                                                                                                                                             |                   |                                                                                                                                                                                                                                                                                                                               |                                                                                 |    |     |     |    |
|-------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------|----|-----|-----|----|
|             |                                                                                                                                                           |                                                                                                                                                                                                             |                   | <p>or manage/supervise in a health system.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Program managers</li> </ul>                                                                                                                                                                    | transparent and effective decisions.                                            |    |     |     |    |
| Tricco 2020 | <p>Commentary</p> <p>Based on author experience conducting COVID-19 rapid reviews predominantly for health care providers and policy decision-makers.</p> | To outline several methodological challenges to the conduct of rapid reviews that have become apparent during the COVID-19 pandemic using an 8-step framework that follows the knowledge synthesis process. | Rapid reviews     | <p>“Stakeholders” without further definition</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Policymakers</li> <li>• Providers of care</li> <li>• Patients</li> </ul>                                                                                                                                             | “Involvement” without further definition                                        | No | No  | Yes | No |
| Troya 2019  | <p>Case study</p> <p>n=3 participants</p> <p>UK</p>                                                                                                       | To critically reflect on the process, potential impact and identify challenges and opportunities in involving robust PPIE in a doctoral systematic review and qualitative study.                            | Systematic review | <p>Patients and the public with previous lived experience with self-harm as an older adult (60 years or older) and/or have worked with (eg support worker) or cared for (eg carer) an older adult with self-harm behaviour.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> </ul> | “Patient Public Involvement and Engagement (PPIE)”, without further definition. | No | Yes | Yes | No |

|             |                                                                                                                                                                                    |                                                                                                                                                                                                                                               |                                     |                                                                                                                                                                                                           |                                                                                                                                         |     |     |     |     |
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| Vale 2012   | Case study and qualitative impact evaluation using questionnaires<br><br>n= 3 patient partners and n=2 researchers<br><br>UK                                                       | To evaluate patient involvement in a systematic review and meta-analysis, from the researcher and patient partner perspective.                                                                                                                | Systematic review and meta-analysis | “Patient Research Partners” were women who had experienced treatment for cervical cancer.<br><br>Groups:<br>● Patients                                                                                    | “Involvement” not explicitly defined; engaged at the level of consultation throughout all steps of the research and evaluation process. | No  | Yes | Yes | No  |
| Wale 2010   | Case study of the Cochrane Consumer Network and two cross-sectional evaluations<br><br>2006: n= 63 consumers and 23 review groups<br><br>2009 n= 66 consumers and 47 review groups | To present what has been accomplished through the involvement of consumers and the Cochrane Consumer Network in the Cochrane Collaboration. The practical and methodological successes, challenges, and practical implications are addressed. | Cochrane systematic reviews         | A consumer is considered a receiver or user of a health service, patient, citizen, carer/caregiver, or layperson.<br><br>Groups:<br>● Patients<br>● Public                                                | Involvement” without further definition.                                                                                                | No  | Yes | Yes | No  |
| Walker 2021 | Case study from the perspective of the research team<br><br>N = 8 young people and 5 parents                                                                                       | To describe and reflect on the methods and influence of involvement of young people with lived experience within a complex evidence synthesis.                                                                                                | Mixed method systematic review      | “Patients” are people with experience of illnesses or conditions that require them to access health services<br>“Public” refers to parents and carers, or people interested in the intersection of mental | Patient and public involvement and engagement (PPIE) in research                                                                        | Yes | Yes | Yes | Yes |



|            |                                                                                       |                                                                                                                                                                                      |                                                                                                                                 |                                                                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                                     |     |     |     |     |
|------------|---------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----|-----|-----|-----|
|            |                                                                                       |                                                                                                                                                                                      |                                                                                                                                 | <p>health and physical long-term conditions for children and young people.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> </ul>                                                              |                                                                                                                                                                                                                                                                                                                                                                     |     |     |     |     |
| Wang 2023  | <p>Scoping review</p> <p>n=9 included studies</p> <p>Up to January 2021</p>           | <p>To describe what activities youth with neurodisabilities and their families have been engaged in as part of evidence syntheses and what were the outcomes of that engagement.</p> | <p>Evidence synthesis including reviews such as, but not limited to, systematic reviews, scoping reviews and meta-analyses.</p> | <p>“Stakeholders” are described as patients, families, health-care providers or policy makers. This paper focuses on patient and family engagement.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> </ul>       | <p>Engagement in research is defined as participation in the research process beyond a participant or ‘subject’ capacity and aims to improve research outcomes by integrating experiences in all aspects of the research. This term is conceptually synonymous with terms such as ‘patient engagement’, ‘patient-oriented research’ and ‘service involvement’ .</p> | Yes | Yes | Yes | Yes |
| Welch 2022 | <p>Systematic methodology review</p> <p>Up to 2021</p> <p>n= 158 included studies</p> | <p>To explore what methods systematic reviewers use to consider health equity in systematic reviews of effectiveness</p>                                                             | <p>Systematic reviews</p>                                                                                                       | <p>“Stakeholders”, persons with lived experience of inequities. Uses the 10P framework to categorize stakeholders.</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Public</li> <li>• Providers</li> </ul> | <p>“Engagement” without further definition</p>                                                                                                                                                                                                                                                                                                                      | No  | No  | No  | Yes |

|               |                                                                                                                                     |                                                                                                                                                                                                                 |                                              |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |                                                                           |     |    |     |    |
|---------------|-------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------|-----|----|-----|----|
|               |                                                                                                                                     |                                                                                                                                                                                                                 |                                              | <ul style="list-style-type: none"> <li>• Principal Investigators</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Payers of health research</li> <li>• Purchasers of health services</li> <li>• Program managers</li> <li>• Peer review editors</li> </ul>                                                                                                                                                                                                                                                     |                                                                           |     |    |     |    |
| Whitlock 2010 | Organizational case study of the Agency for Healthcare Research and Quality's (AHRQ) Effective Health Care (EHC) program<br><br>USA | To discuss the identification, selection, and refinement of topics for comparative effectiveness systematic reviews within the Agency for Healthcare Research and Quality's Effective Health Care (EHC) program | Comparative effectiveness systematic reviews | <p>"Stakeholders" without further definition. Categories for the Effective Health care program include Clinicians, consumers/patients, employers and business groups, federal and state partners, healthcare industry representatives, payers, health plans, policy makers, and researchers</p> <p>Groups:</p> <ul style="list-style-type: none"> <li>• Patients</li> <li>• Providers</li> <li>• Principal Investigators</li> <li>• Policymakers</li> <li>• Product makers</li> <li>• Payers of health research</li> </ul> | Referred to as "engagement" and "involvement" without further definition. | Yes | No | Yes | No |

|  |  |  |  |                                                                                                             |  |  |  |  |  |
|--|--|--|--|-------------------------------------------------------------------------------------------------------------|--|--|--|--|--|
|  |  |  |  | <ul style="list-style-type: none"><li>• Purchasers of health services</li><li>• Providers of care</li></ul> |  |  |  |  |  |
|--|--|--|--|-------------------------------------------------------------------------------------------------------------|--|--|--|--|--|

## Supplemental 5: Equity considerations across PROGRESS-Plus characteristics

| Equity characteristics                                        | Considerations for engagement                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
|---------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Place of residence<br>(n=4 studies)                           | <p><i>High income vs. low- and middle-income country engagement</i></p> <ul style="list-style-type: none"> <li>• Consumer involvement in the Cochrane Collaboration is overwhelmingly in developed countries: two-thirds were from the UK and North America with others in Australia and Europe and only 4/63 from Asia or the Middle East. Data from Cochrane Review Group case studies report similar patterns, suggesting barriers to engagement exist in developing countries, such as information scarcity, education, funding, communication system difficulties and lack of facilities [Morley 2016].</li> <li>• Priority setting for Cochrane systematic reviews has reportedly been conducted among predominantly White people from the UK, and a lack of diversity may have led to a biased set of priorities. To address this, topics which were prioritized by people originating from low- and middle-countries can be intentionally retained [Dennett 2022]. Hosting workshops online makes it possible to attract a wider variety of international interest-holders and give rise to valuable discussions on the differences in health systems and models of care [O'Connor 2023].</li> <li>• Engaging interest-holders in multiple steps of the review priority-setting process can help promote evidence-informed policy-making and practice by aligning systematic review production to policy priorities and channeling limited resources to areas of highest priority. This is particularly relevant in the context of low- and middle-income countries, where the capacity for producing systematic reviews is limited and often misaligned with policy needs and priorities [Fadlallah 2020].</li> </ul> |
| Race, ethnicity, culture, language, ancestry<br>(n=8 studies) | <p><i>Language and cultural differences</i></p> <ul style="list-style-type: none"> <li>• Patient authors may provide stronger contributions if able to provide input in their local language [Arnstein 2020].</li> <li>• Tools such as the TRANSFER conversation guide can facilitate engagement, but it was noted that it would be helpful to have a translation of the conversation guide available in an interest-holders' own language [Munthe-Kaas 2022].</li> <li>• Patients need more support in understanding terminology and research processes when English is not their primary language [Bayliss 2016].</li> <li>• Cochrane review groups operating in non-Western countries (e.g. China) experience barriers to consumer involvement including language and cultural differences [Morley 2016].</li> <li>• The existing Cochrane Consumer Executive faces an overwhelming challenge in supporting consumer involvement within the work of Cochrane across the globe. The challenges to effective support for consumer involvement across the network include language and cultural differences [Cochrane Collaboration, 2015].</li> </ul> <p><i>Reporting of language/race/ethnicity</i></p> <ul style="list-style-type: none"> <li>• Key informants in a US context were 100% White, with no Hispanics/Latinos or other Non-White groups represented [Cornman 2017]</li> <li>• We used a sampling frame to guide workshop recruitment and approached specific organizations representing groups in</li> </ul>                                                                                                                                                                                                    |

|                                        |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                          |
|----------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|                                        | <p>the recruitment process, but participation rates in the workshop remained low (3/ 28 people from culturally and linguistically diverse backgrounds and 1/28 Indigenous people) [Synnot 2020]</p> <ul style="list-style-type: none"> <li>• All patient/public members were of a white-British background, limiting the voice of minority groups in the research [Troya 2019].</li> </ul>                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                               |
| Occupation<br>(n=3 studies)            | <ul style="list-style-type: none"> <li>• At a collaborative level, a parent of children with ADHD with experience of working as a teaching assistant and running a support group for parents of children with ADHD and a behavioural support professional were an integral part of the project team. [Coon 2016]</li> <li>• Roger is a retired mental health social worker who lives with lymphoma (in long-term remission) and heart failure. [Hannigan 2022]</li> <li>• consumer or carer representative, with roles including consumer and community advisory committee members, board members and voluntary office bearers in organizations such as health services, government departments or agencies, not-for-profit organizations and charities. [Synnot 2019]</li> </ul>                                                                                                                                                                                                                                                                                                                                        |
| Gender/Sex<br>(n=2 studies)            | <p><i>Gender or sexual minorities are hidden</i></p> <ul style="list-style-type: none"> <li>• It was not possible to recruit any patient/public members belonging to lesbian, gay, bisexual and transsexual (LGBT) groups, who also engage in self-harm behavior which is often hidden. Patient/public members in this research were all of a white-British heterosexual background, limiting the voice of minority groups in the research [Troya 2019].</li> <li>• In the case of the review about HIV health promotion for men who have sex with men, the researchers expressed a concern that the public who took active part in the review process were drawn exclusively from members of organizations representing the interests of men who have sex with men, which could be judged to have excluded men who have sex with men but who do not have such affiliation [Boote 2011]</li> </ul>                                                                                                                                                                                                                       |
| Religion<br>(n=0 studies)              | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |
| Education<br>(n=0 studies)             | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |
| Socio-economic status<br>(n=3 studies) | <p><i>Access to technology and resource bias</i></p> <ul style="list-style-type: none"> <li>• Interest-holders with lower access to technology may not have the ability to respond to invitations to engage in reviews or their resources may be too limited to allow full engagement throughout the systematic review process, producing a biased, unbalanced group. Review teams should minimize necessary resources needed for engagement, for example by reducing unnecessary reading. Interest-holders may not be able to attend physical meetings due to limited time or money. Providing stakeholders with multiple modes of interaction (for example workshops, email, face-to-face interviews, etc.) can mitigate access/technology bias and resource bias. Resources to aid travel to physical meetings can also be provided [Haddaway 2017].</li> <li>• Online engagement is generally considered positive overall, but many individuals recommended having at least one in-person meeting (per year) during a project, as online-only engagement may ‘miss the seldom seen voices’ highlighting a</li> </ul> |

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|------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|                                    | <p>need to engage with ‘different and wider communities’ [Gavin 2023].</p> <ul style="list-style-type: none"> <li>• Online-only recruitment can limit the diversity in interest-holders that are invited to engage, potentially engaging those who have access to these types of opportunities (e.g. ‘super patients’ – people who are included more often and can dedicate more time). Efforts need to be made to engage interest-holders with diverse backgrounds and experiences (e.g. income and education) [Wang 2023].</li> </ul>                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                    |
| Social capital                     | None reported                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                              |
| Age (plus)<br>(n=3 studies)        | <p><i>Representation</i></p> <ul style="list-style-type: none"> <li>• Empowering stakeholder voices is particularly important when it comes to those of young people, who are typically underrepresented within a literature base which often lacks the depth of exploration required to explore generative causal factors [Cooper 2020].</li> <li>• The researchers reiterated children and young people were the only individuals in the team able to provide a perspective on what it is like to be a young person dealing with a long-term physical condition and mental health issues and their experiences were invaluable to success, since that expertise was critical to the research [Walker 2020].</li> </ul> <p><i>Caregiver consent</i></p> <ul style="list-style-type: none"> <li>• Including interest-holders under 16 years of age required informed consent from them and their caregiver (in addition to ethical approval), which can add a level of administration often not required in realist syntheses [Cooper 2020].</li> </ul> <p><i>Age-appropriate language</i></p> <ul style="list-style-type: none"> <li>• Care should be taken to ensure that the language used during discussion is age-appropriate [Walker 2020].</li> <li>• Some individuals [service users] questioned their own authority, ability or worthiness to represent their organization. One person told us they were interested but feared they were ‘too old’; another person described himself as being ‘too stupid’. In these instances, we reassured potential participants [...] such reassurance could be perceived as being patronizing (Smith 2009).</li> </ul> <p><i>Co-production considerations</i></p> <ul style="list-style-type: none"> <li>• It is not clear whether children and young people and their parents would welcome the level of responsibility associated with a co-produced research synthesis (e.g., establishment of a working relationship and sharing of research tasks prior to project conception, through grant application, conducting and writing up the project and dissemination). A blended model of co-production and consultation may be more appropriate, to allow each interest-holder to use their relevant skills and expertise to fulfill the demands of the project [Walker 2020].</li> </ul> |
| Disability (plus)<br>(n=4 studies) | <p><i>Facilitators for engagement</i></p> <ul style="list-style-type: none"> <li>• Youth with neurodisabilities and their families were often recruited from established organizations and social networks. Online social media platforms such as Facebook are beneficial for building research partnerships because they can connect a wide range of interest-holders. Closed Facebook groups like Patients Partnering in Research enable researchers, trainees, and patients and families to build connections and facilitate recruitment in a virtual space. [Wang</li> </ul>                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                           |

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|--|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|  | <p>2023]</p> <ul style="list-style-type: none"> <li>Review authors hosted a workshop and extended a research training developed for patients with mental disorders, including information about systematic reviews and meta-analyses in the context of evidence-based medicine (Brutt 2017)</li> </ul> <p><i>Sensitivity to health conditions</i></p> <ul style="list-style-type: none"> <li>Care was needed to ensure that discussing their experiences did not cause the mental or physical health of children and young people to worsen or mean that attending meetings was experienced as similar to attending a personal therapy session [Walker 2020]</li> <li>If children and young people were unable to attend meetings due to poor mental or physical health, a member of the team sent them a summary of what was discussed and invited them to give their own feedback over the telephone or via email. The team made it clear that being unable to attend a meeting in person would not exclude children and young people from attending future meetings, as they understood that their health experiences could vary across the 15-month project. The incorporation of a dedicated patient and public involvement co-investigator was essential in facilitating flexible involvement [Walker 2020].</li> </ul> <p><i>Representativeness</i></p> <ul style="list-style-type: none"> <li>Patients were invited to participate on the basis of electronic inter-communication from the heads of patient associations, so it is impossible to know whether our participants are sufficiently representative of patients with degenerative ataxias as a whole [Boote 2011]</li> </ul> |
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## **CHAPTER 4: ARTICLE 2. Barriers and facilitators to stakeholder engagement in health guideline development: A qualitative evidence synthesis**

### **Chapter Overview**

This Chapter presents a qualitative evidence synthesis on barriers and facilitators to engagement in health guideline development. It has been submitted to *Campbell Systematic Reviews* for publication. To note, this Chapter uses the term “stakeholder”, as this was the term used by the MuSE Consortium at the time of writing. This will be updated to “interest-holder” during peer review revisions.

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### **Contributions (CRediT):**

**Conceptualization:** O Magwood, AY Riddle, J Petkovic, J Khabsa, L Lytvyn, P Campbell, TW Concannon, EA Akl, V Welch, P Tugwell

**Methodology:** O Magwood, AY Riddle, J Petkovic, J Khabsa, L Lytvyn, P Campbell, TW Concannon, EA Akl, V Welch, P Tugwell

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**Project administration:** O Magwood, J Petkovic

**Funding acquisition:** P. Tugwell, P. Campbell, TW Concannon, L Dans, J Petkovic, V Welch

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**Declarations of interest:** O Magwood, S Grant, P Tugwell, TW Concannon, EA Akl, and V Welch are co-authors of studies which have been included in this review. Authors of included studies were not involved in the decision to include, critical appraisal or data extraction of studies on which they were a coauthor.

V Welch is Editor-in-Chief of the Campbell Collaboration and was not involved in editorial decisions regarding this review.

P Tugwell provides independent medical consultation professional services to the Reformulary Group . He is [unpaid] Chair of the Management Group of a registered non-profit independent medical research organization, OMERACT, whose goal is to improve and advance the health

outcomes for patients suffering from musculoskeletal conditions. OMERACT receives arms-length funding from 11 companies (Abbvie, Astra Zenaca, Aurinia, BMS, Centrexion, GSK, Horizon Pharma Inc , Janssen, Novartis, Pfizer & Sparrow).

S Grant's spouse is a salaried employee of, and owns stock in, Eli Lilly and Company. SG has accompanied his spouse on company-sponsored travel.

S Kumbargere Nagraj is a Senior Researchers with the MAGIC Evidence Ecosystem Foundation, which is involved in the production of clinical practice guidelines. His work included drafting and producing CPGs. The results and conclusion of this research work might be referred in future work of MAGIC. The majority of work done in this review was before joining the MAGIC foundation.

The other authors declare no conflicts of interest.

## **4.1 ABSTRACT**

### **Background**

Health guidelines are important resources for informing decisions made by health practitioners, patients, caregivers, policymakers, and other stakeholders. Stakeholder engagement in health guideline development can improve the relevance of recommendations, promote considerations of equity, and support implementation. To facilitate efficient and effective health guideline development, there is a need for a comprehensive, global, evidence-based guidance for stakeholder engagement in guideline development. This paper synthesises the barriers and facilitators to stakeholder engagement in guideline development. Several systematic reviews have examined the topic of stakeholder engagement in guideline development, but few have explicitly focused on barriers and facilitators to engagement and the majority of these were related to patients. With increased recognition of the value of broad stakeholder engagement, understanding the barriers and facilitators to stakeholder engagement is key to developing a relevant and inclusive health guideline.

### **Objectives**

The objective of this review is to identify and synthesise the reported evidence on barriers and facilitators to stakeholder engagement in health guideline development. We address this objective through two research questions:

1. What are the reported barriers to multi-stakeholder engagement in health guideline development across the 18 topics of the GIN-McMaster checklist?
2. What are the reported facilitators to multi-stakeholder engagement in health guideline development across the 18 topics of the GIN-McMaster checklist?

### **Search methods**

We searched five major electronic databases (MEDLINE (OVID), Cumulative Index to Nursing & Allied Health Literature (CINAHL; EBSCO), EMBASE (OVID), PsycInfo (OVID), Scopus and Sociological Abstracts). We also conducted an extensive grey literature search using the websites of agencies who actively engage stakeholders in research and/or guideline-producing agencies, such as PCORI, WHO and GIN. We searched from database inception up to the 26th September, 2022. Backward and forward citation tracking was performed on included articles to identify other eligible studies.

### **Selection criteria**

We included primary research studies which qualitatively reported on the barriers or facilitators to stakeholder engagement in health guideline development. This included qualitative or mixed method research studies using methods such as interviews, focus groups, or surveys to collect participant experiences; case studies of existing programmes; and process evaluation studies. We excluded non-empirical publications including commentaries and editorials. We excluded publications with incomplete data, including conference abstracts and protocols.

We defined stakeholders as “any individual or group who is responsible for or affected by health- and healthcare-related decisions”. We identified 10 types of stakeholders whose input can enhance the relevance and uptake of guidelines. We included studies that reported on the barriers or facilitators to engagement of one or several of these stakeholder groups at any step of the guideline development process.

### **Data collection and analysis**

All identified citations from electronic databases were imported into Covidence for screening and selection. Documents identified through our grey literature search were managed and

screened using an Excel spreadsheet. A two-part study selection process was used for all identified citations: (1) a title and abstract review and (2) full-text review. At each stage, teams of two review authors independently assessed all potential studies in duplicate using a priori inclusion and exclusion criteria.

Data was extracted from each included article in duplicate and independently. We extracted information about study characteristics and methods. Additionally, we extracted qualitatively reported barriers and facilitators and conducted a framework analysis. We selected the Theoretical Domains Framework (TDF) for extracting our barriers and facilitator data.

Developed through a multidisciplinary consensus approach and subsequent validation, TDF consolidates overlapping behavioural theories into 14 domains encompassing 84 theoretical constructs, and provides a theoretical lens through which to view the cognitive, affective, social and environmental influences on behaviour and implementation. Two review authors conducted the analysis.

## **Main results**

Our qualitative evidence synthesis identified a total of 52 findings, encompassing 24 barriers and 28 facilitators to stakeholder engagement in health guideline development. While the large majority of reported evidence is about patient engagement, many findings are applicable to all stakeholder groups and should be considered by guideline developers who aim to apply a multi-stakeholder engagement approach.

Many organisations that produce guidelines lack the resources to support stakeholder engagement. By implementing an organisational stakeholder engagement model, guideline developers can ensure that they have the necessary human and financial resources in place. The stakeholders which are recruited to a guideline project should be knowledgeable and experienced, and guideline developers should consider diverse perspectives, roles, and personal characteristics. Using a network approach for identification and recruitment can help identify appropriate and committed stakeholders. To prevent tokenistic engagement, guideline developers can promote the meaningful engagement of stakeholders by providing practical, technical and

emotional support. This includes providing training to empower stakeholders and improve self-efficacy and confidence in guideline development skills. Training should focus on evidence-based methods such as systematic reviews and evidence-to-decision frameworks.

Engagement throughout all stages of guideline development supports more meaningful engagement compared to occasional involvement. A skilled moderator/chair can manage group dynamics and support the contributions of all those involved in guideline development. They can also offer explanations and promote plain language as-needed. Finally, many stakeholders have limited time to contribute to guidelines, and developers should establish flexible processes and timelines, and consider stakeholders preferences for in-person and online engagement activities.

### **Authors' conclusions**

Identifying and analysing barriers and facilitators to stakeholder engagement is important in order to optimise health guideline development. Knowing facilitators to engagement can support future multi-stakeholder work. More research is needed on the barriers that do not have potential facilitator solutions, to understand how to improve upon known challenges. This review calls for the development of training courses and tools to support and facilitate involvement of all stakeholders, both public and professionals. In addition, it calls for further research to be done with a focus on the dynamics of guideline development, perceived interactions within the group and their impact on decisions. Work should also be done to look at the implementation of guidelines and the influence of multi-stakeholder engagement on uptake.

## **4.2 PLAIN LANGUAGE SUMMARY: What challenges do patients and other stakeholders face when creating health guidelines, and how can we support them?**

### **The review in brief:**

- Health guidelines help many people, such as clinicians and patients, make decisions about healthcare. It is necessary to involve different types of people in the creation of health guidelines.
- This review summarises the challenges that people face when working together to create health

guidelines and what kind of support helps them work together.

- Most of what we know about the experience of partnership when developing health guidelines is from patients.
- The main challenges facing patients are unclear roles, need for training and support, difficulty speaking up, and not having enough time to do the work. To support them, guideline developers should offer practical, technical and emotional support and work with flexible timelines and processes.
- More research is needed to understand the challenges faced by policymakers, practitioners, researchers and patients from different backgrounds and experiences.

### **What challenges do patients and other stakeholders face when creating health guidelines, and how can we support them?**

Health guidelines are important documents that can help people, such as practitioners and patients, make decisions about healthcare. It is becoming more common to involve different types of people in the creation of health guidelines. We wanted to know what challenges people face when working together to create health guidelines, and what types of supports are useful.

### **What did we do?**

We looked for literature that described people's experiences when collaborating to create health guidelines. We specifically looked for information that described challenges they faced, how they overcame these challenges, and what type of supports they wished were in place. We were interested in documents that described ten different types of people who can contribute to the guideline development process, including patients, practitioners, and members of the public. We organised our results using a pre-existing checklist (GIN-McMaster Checklist for Guideline Development).

### **What did we find?**

We found 40 documents that described challenges and ways to support people who are engaged

in the guideline development process. Most of the evidence came from high-income countries and most of it was about patients and their experience being part of guideline development. These documents described how finding the right people to include in a guideline project can be challenging, since they need specific skills and knowledge or experience. People described not always understanding their role in the project, and needing more training and support in order to do their work. Organisations that develop guidelines did not always have the resources needed to support them. An important resource is the guideline chair, who leads the group discussions and can support everyone involved in creating the health guideline. The best way to support people is to provide them with choices and flexibility in how they engage in the guideline development process. People may prefer working in-person or online and need enough time to get the work done.

### **What are the limitations of the evidence?**

Most of the evidence we found is about patients, and more research is needed about different stakeholders such as policy makers, health practitioners, and researchers. More information is also needed about how to involve people from diverse backgrounds, such as those who live in low-income settings. Additionally, we did not find evidence for all of the steps needed to create a guideline, such as priority setting.

### **How up-to-date is this evidence?**

Evidence was included up to September 26, 2022.

## **4.3 BACKGROUND**

### **The problem, condition or issue**

Health guidelines are “systematically developed evidence-based statements which assist providers, recipients and other stakeholders in making informed decisions about appropriate health interventions” (WHO 2003). Guidelines play a crucial role in the delivery of evidence-based medicine. The systematic examination of evidence conducted through a comprehensive search of the literature, critical appraisal of the quality of the evidence and interpretation of the



findings in light of patients’ preferences and societal values (Guyatt 2008). These guidelines assist health practitioners, patients, caregivers, policymakers, and other stakeholders to make informed decisions about health practice, public health and health policy. It is increasingly recognised that the engagement of multiple stakeholders in guideline development improves recommendation relevancy, uptake, implementation and sustainability (Moulding 1999; Gagliardi 2012; Esmail 2015).

Stakeholders are "any individual or group who is responsible for or affected by health- and healthcare-related decisions that can be informed by research evidence" (Concannon 2012). Our team previously reviewed a number of frameworks for categorising stakeholder groups in research (Concannon 2012; Concannon 2019) and developed a ‘10P’s’ framework for stakeholders in guideline development (Petkovic 2023) [See Table 1].

**Table 1.** Stakeholder categories and their descriptions (Petkovic 2023)

| Stakeholder category                                            | Description                                                                                                                                                                                                  |
|-----------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Patients, patient caregivers, patient advocates/organisations   | Those with lived experience with the condition of interest or who care for or advocate on behalf of those with lived experience                                                                              |
| Payers of health research                                       | Individuals and organisations that fund research projects, such as government funders, industry funders, foundations                                                                                         |
| Payers / Purchasers of Health Services                          | Individuals, organisations and entities that pay for health services                                                                                                                                         |
| Peer Reviewed Journals Editors                                  | Those who set journal policy on guidelines and manage the peer review process and editing                                                                                                                    |
| Policymakers                                                    | Individuals, organisations and entities that craft public or private policy (on health) at any level of government                                                                                           |
| Principal Investigators and all members of the of research team | Individuals, organisations, and associations that conduct or advocate health research                                                                                                                        |
| Product makers                                                  | Individuals working for companies that manufacture pharmaceuticals, medical devices, medical procedures, health technologies, or for profit educational and behavioural packages                             |
| Program managers                                                | Managers/directors who plan, lead, oversee, or deliver any program that provides public health, community services, or clinical care (e.g. budgeting, hiring, staffing, organising, coordinating, reporting) |
| Providers                                                       | Persons and their professional associations who provide health care in a professional capacity and allowed by regulatory bodies to provide a health care service                                             |
| Public                                                          | Individuals in the general population of a defined geographic area excluding patients, caregivers, and health professionals living or working with the condition of interest                                 |

Major guideline developers, including the World Health Organization (WHO), the Canadian

Institute for Health Research (CIHR) and the National Institute for Health and Care Excellence (NICE), as well as the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) Working Group recommend the inclusion of stakeholder groups in the guideline development and implementation process (WHO 2003; CIHR 2014; NICE 2014; Akl 2017). They recommend this involvement to improve considerations of equity (Akl 2017) and to ensure that the final guideline materials are useful and understandable for its end-users (WHO 2014). However, it is not clear how best to engage stakeholders or at what stage of the guideline development cycle. There is a need for the development of comprehensive, global, evidence-based guidance for stakeholder engagement in health guideline development that brings together the evidence (literature and stakeholder expertise) amassed to date.

The Multi-Stakeholder Engagement Consortium (MuSE Consortium) was established in 2015 to advance methods and approaches used in stakeholder engaged health research. The consortium is focused on engagement in health research and practice – including reviews and guidelines -- and includes more than 140 researchers, trainees and stakeholders from over 20 countries (Petkovic 2020). As part of the consortium's work, a working group was formed to develop global guidance on stakeholder engagement in health guideline development. The working group set out to conduct four concurrent systematic reviews to summarise the evidence on: (1) existing guidance for stakeholder engagement in guideline development, (2) barriers and facilitators to stakeholder engagement in guideline development, (3) managing conflicts of interest in stakeholder engagement in guideline development, and (4) measuring the impact of stakeholder engagement in guideline development. This review is the second in this series.

### **Description of the phenomena of interest**

For the purpose of this review, we define **guidelines** as “systematically developed evidence-based statements which assist providers, recipients and other stakeholders in making informed decisions about appropriate health interventions” (WHO 2003). The steps to consider for guideline development are outlined by the GIN-McMaster Checklist (Schunemann 2014), a comprehensive checklist of items that guideline developers could consider for all stages of the guideline enterprise, from the planning and formulation of recommendations, to their

implementation, evaluation and updating. The checklist comprises 18 overarching topics, including “Consumer and stakeholder involvement” (topic 6) [See Table 2].

**Table 2:** Topics included in the GIN-McMaster checklist for guideline development

| Topic                                              | Description                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |
|----------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| 1. Organisation, budget, planning and training     | Involves laying out a general but detailed plan describing what is feasible, how it will be achieved and what resources are required to produce and use the guideline. The plan should refer to a specific period and be expressed in formal, measurable terms.                                                                                                                                                                                                                                                                        |
| 2. Priority setting                                | Refers to the identification, balancing and ranking of priorities by stakeholders. Priority setting ensures that resources and attention are devoted to those general areas (e.g., chronic obstructive pulmonary disease, diabetes, cardiovascular disease, cancer, prevention) where health care recommendations will provide the greatest benefit to the population, a jurisdiction or a country. A priority-setting approach needs to contribute to future plans while responding to existing, potentially difficult circumstances. |
| 3. Guideline group membership                      | Defines who is involved, in what capacity, and how the members are selected for the guideline development and at other steps of the guideline enterprise.                                                                                                                                                                                                                                                                                                                                                                              |
| 4. Establishing guideline group processes          | Defines the steps to be followed, how those involved will interact and how decisions will be made.                                                                                                                                                                                                                                                                                                                                                                                                                                     |
| 5. Identifying target audience and topic selection | Involves describing the potential users or consumers of the guideline and defining the topics to be covered in the guideline (e.g., diagnosis of chronic obstructive pulmonary disease).                                                                                                                                                                                                                                                                                                                                               |
| 6. Consumer and stakeholder involvement            | Describes how relevant people or groups who are not necessarily members of the panel but are affected by the guideline (e.g., as target audience or users) will be engaged.                                                                                                                                                                                                                                                                                                                                                            |

|                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                        |
|--------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| 7. Conflict of interest considerations                                                     | Focuses on defining and managing the potential divergence between an individual's interests and his or her professional obligations that could lead to questioning whether the actions or decisions are motivated by gain, such as financial, academic advancement, clinical revenue streams or community standing. Financial or intellectual or other relationships that may affect an individual's or organisation's ability to approach a scientific question with an open mind are included.                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                       |
| 8. Question generation                                                                     | Focuses on defining key questions the recommendations should address using the PICO (patient/problem, intervention, comparison, outcome) framework, including the detailed population, intervention (including diagnostic tests and strategies) and outcomes that will be relevant for decision-making (e.g., should test A be used, or should treatments B, C, D or E be used in chronic obstructive pulmonary disease?).                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                             |
| 9. Considering importance of outcomes and interventions, values, preferences and utilities | Includes integrating, in the process of developing the guidelines, how those affected by its recommendations assess the possible consequences. These include patient, caregiver and health care provider knowledge, attitudes, expectations, moral and ethical values, and beliefs; patient goals for life and health; prior experience with the intervention and the condition; symptom experience (e.g., breathlessness, pain, dyspnea, weight loss); preferences for and importance of desirable and undesirable outcomes; perceived impact of the condition or interventions on quality of life, well-being or satisfaction, and interactions between the work of implementing the intervention, the intervention itself, and other contexts the patient may be experiencing; preferences for alternative courses of action; and preferences relating to communication content and styles, information and involvement in decision-making and care. This can be related to what in the economic literature is considered <i>utilities</i> . An intervention itself can be considered a consequence of a recommendation (e.g., the burden of taking a medication or undergoing surgery) and a level of importance or value is associated with that. |
| 10. Deciding what evidence to include and searching for evidence                           | Focuses on laying out inclusion and exclusion criteria based on types of evidence (e.g., rigorous research, informally collected), study designs, characteristics of the population, interventions and comparators, and deciding how the evidence will be identified and obtained. It also includes but is not limited to evidence about values and preferences, local data and resources.                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                             |
| 11. Summarising evidence and considering additional information                            | Focuses on presenting evidence in a synthetic format (e.g., tables or brief narratives) to facilitate the development and understanding of                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                             |

|                                                                                                   |                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |
|---------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|                                                                                                   | recommendations. It also involves identifying and considering additional information relevant to the question under consideration.                                                                                                                                                                                                                                                                                                                                         |
| 12. Judging quality, strength or certainty of a body of evidence                                  | Includes assessing the confidence one can place in the obtained evidence by transparently evaluating the obtained research (individual studies and across studies) and other evidence applying structured approaches. This may include, but is not limited to, evidence about baseline risk or burden of disease, importance of outcomes and interventions, values, preferences and utilities, resource use (cost), estimates of effects and accuracy of diagnostic tests. |
| 13. Developing recommendations and determining their strength                                     | Developing recommendations involves use of a structured analytic framework and a transparent and systematic process to integrate the factors that influence a recommendation. Determining the strength of the recommendations refers to judgments about how confident a guideline panel is that the implementation of a recommendation exerts more desirable than undesirable consequences.                                                                                |
| 14. Wording of recommendations and of considerations about implementation, feasibility and equity | Refers to choosing syntax and formulations that facilitate understanding and implementation of the recommendations. Such wording is connected to considerations about implementation, feasibility and equity, which refer to the guideline panel's considerations about how the recommendation will be used and what impact it may have on the factors described.                                                                                                          |
| 15. Reporting and peer review                                                                     | Reporting refers to how a guideline will be made public (e.g., print, online). Peer review refers to how the guideline document will be reviewed before its publication and how it can be assessed (e.g., for errors), both internally and externally, by stakeholders who were not members of the guideline development group.                                                                                                                                            |
| 16. Dissemination and implementation                                                              | Focuses on strategies to make relevant groups aware of the guidelines and to enhance their uptake (e.g., publications and tools such as mobile applications).                                                                                                                                                                                                                                                                                                              |

|                        |                                                                                                                                                                                                                                                                                                             |
|------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| 17. Evaluation and use | Refers to formal and informal strategies that allow judgments about: evaluation of the guidelines as a process and product; evaluation of the use or uptake, or both; and evaluation of impact and whether or not the guideline leads to improvement in patient or population health or other consequences. |
| 18. Updating           | Refers to how and when a guideline requires revision because of changes in the evidence or other factors that influence the recommendations                                                                                                                                                                 |

Further, we define **stakeholders** as "any individual or group who is responsible for or affected by health- and healthcare-related decisions that may be informed by research evidence" (Concannon 2012). We recognize a number of negative historical uses of the word "stakeholder", such as its "mercenary" roots as the person who physically held the stakes in a bet (Sharfstein 2016) or its colonial roots in which a stakeholder was the person who drove a stake into the land to demarcate the land they were occupying/stealing from Indigenous territories. In this context, it has been suggested that stakeholders should instead be referred to as "partners" or "rightsholders" (e.g., Government of British Columbia 2023). Additional terminology has been put forward by Reed and Rudman (2022) who replaced the word "stakeholder" with "interested/affected groups and non-academic partners" (Reed 2023). "Interested/ affected groups" are not directly engaged with the research (and so cannot be referred to as partners), but may still have significant influence (typically individuals and organisations in gatekeeper roles) or may be significantly impacted by research but have limited interest or influence (typically marginalised, powerless and often oppressed groups). However, this terminology does not accurately capture academic partners (e.g., principal investigators, guideline methodologists, clinician-scientists) who have a critical role in guideline development. Therefore, with acknowledgement of some of the problematic history of the term, we use the term 'stakeholder' throughout this review as it encompasses the range of individuals and/or groups we want to include.

In the context of this manuscript, **engagement** is an approach to ensure the contribution of stakeholders towards the development of the guideline, completion of any of the stages of the guideline, or dissemination of the guideline and its recommendations (Frank 2020; Pollock

2018). There are terms that have been used to talk about engagement with specific stakeholders, including “public involvement”, “consumer engagement”, and “Patient and Public Involvement (PPI)”. Other terms can describe engaging in partnership across stakeholder groups, such as “co-production”, “co-creation”, “activation” and “involvement”. A stakeholders’ **level of engagement** is the intensity of involvement in the guideline development process (Oliver 2008; Pollock 2019). Through a review of existing sources and a series of multi- stakeholder discussions, we operationalize two levels of engagement for the purpose of this review (Petkovic 2023). Our first level is ‘advice/feedback’; stakeholder opinions, perspectives, experiences, or values are sought and considered by the guideline development team. Our second level of engagement is ‘decision-making’ which includes collaboration and co- production; stakeholders actively contribute to making decisions at the different stages of the guideline’s development and recommendations. While there are nuances within each of our two levels of engagement, we have decided that the use of two clearly distinct levels is more feasible for the assessment of stakeholder engagement across the topics included in the GIN- McMaster checklist for guideline development. Figure 1 depicts the complex and iterative engagement of stakeholders across these topics.

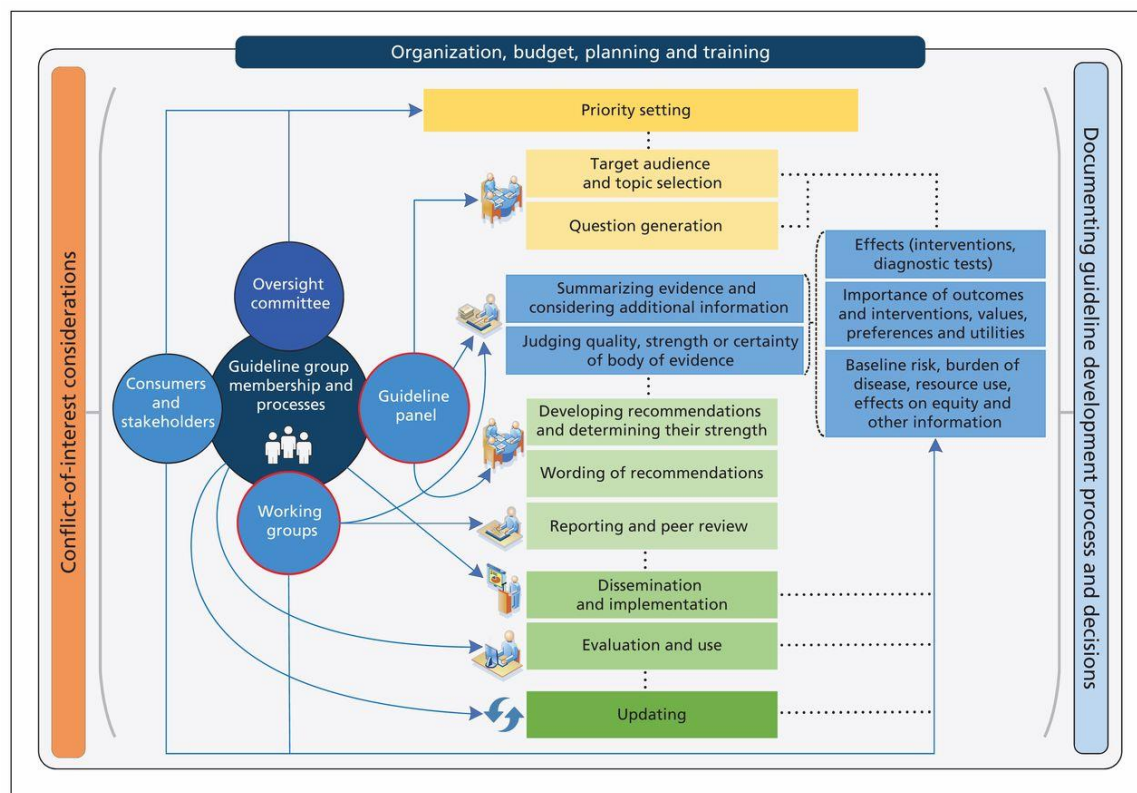


**Figure 1. Diagram of the guideline development process.** “The steps and involvement of various members of the guideline development group are interrelated and not necessarily sequential. The guideline panel and supporting groups (e.g., methodologist, health economist, systematic review team and secretariat for administrative support) work collaboratively, informed through consumer and stakeholder involvement. They typically report to an oversight committee or board overseeing the process. For example, while deciding how to involve stakeholders early for priority setting and topic selection, the guideline group must also consider how developing formal relationships with the stakeholders will enable effective dissemination and implementation to support uptake of the guideline. Furthermore, considerations for organisation, planning and training encompass the entire guideline development project, and steps such as documenting the methodology used and the decisions made, as well as considering conflicts of interest, occur throughout the process” (Schunemann 2014)

Many activities are desirable for effective stakeholder engagement in guideline development (Figure 2). The engagement of stakeholders from multiple disciplines or expertise is typically recommended to take account of a wide range of opinions. Indeed, greater diversity in stakeholders' background increases the knowledge and skills available to make decisions (Oliver 2018). However, the ability to gather and apply input from multiple stakeholders in a way that meaningfully reflects and addresses stakeholders' interests is not easily achieved, as increased diversity may also reduce group cohesiveness (Oliver 2018). In their review of theoretical literature, Brodbeck and colleagues (Brodbeck 2007) proposed that groups can outperform individual decision makers under conditions which promote sharing of knowledge and mutual learning. Oliver and colleagues (Oliver 2018) built on this model, and suggested that these conditions include diverse group membership, skilled facilitation, time for knowledge sharing, following a formal consensus processes, and integrating divergent perspectives.



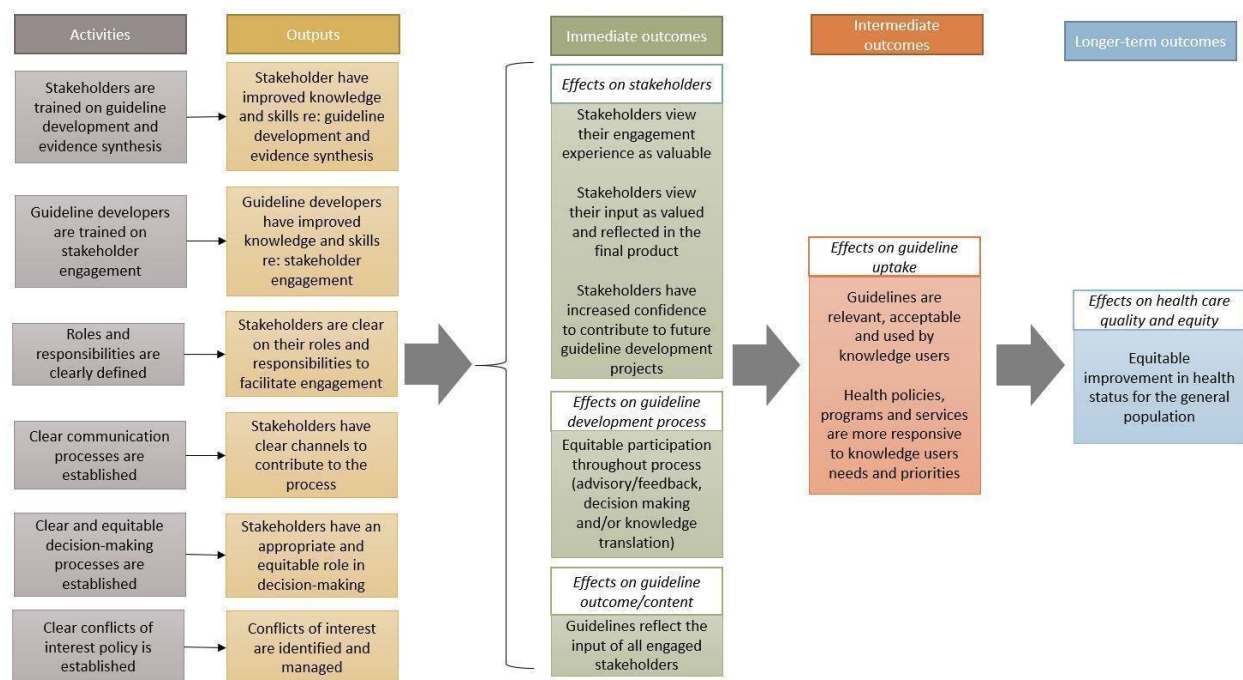


**Figure 2. Logic model of the effects of stakeholder engagement in guideline development**

Stakeholder engagement is a complex process, and successful engagement requires both individual and organisational adoption. Organisational cultures and relationships act as a backdrop to these stakeholder engagement activities (Oliver 2018). There are a multitude of individual- and organisational-level factors that can affect a stakeholder's ability to engage in the guideline development process, or a guideline developer's ability to effectively engage them.

**Barriers** to stakeholder engagement are defined as any variable, condition or factor that may impede stakeholder engagement in guideline development, while **facilitators** are any variable, condition or factor that may promote stakeholder engagement in guideline development. Such determinants may include knowledge, skills, time, and resources, for example, and may occur across one or more topics of guideline development.

### Why it is important to do this review

With increased recognition of the value of stakeholder engagement in guideline development, there is a need for expert opinion and evidence-based guidance on when and how to engage stakeholders. This review of barriers and facilitators to stakeholder engagement in guideline

development will help fill this gap. It will build on existing reviews of barriers and facilitators, synthesize the current evidence-base, and examine a broad range of stakeholder groups.

To date, several systematic reviews have been conducted on the topic of stakeholder engagement in guideline development, but few are explicitly focused on barriers and facilitators to engagement. Oxman and colleagues (Oxman 2006) conducted a series of reviews to advise WHO to ensure that health care recommendations are informed by the best available research evidence. Relevant reviews in this series included group composition (Fretheim 2006a), group processes (Fretheim 2006b), and consumer involvement (Schunemann 2006). However, these reviews were not systematic nor exhaustive, and prioritized existing syntheses over primary empirical studies. Relatedly, Oliver and colleagues (Oliver 2018) synthesized theoretical literature to learn about the structure, processes, and environment of committees generally, which has applications for health guideline development. Given the recent promotion of co-production in health and the rapid pace of involvement of diverse people in all types of research, these reviews are likely out of date and require updating to better capture this evolving landscape.

Much of the existing barrier and facilitator syntheses focus specifically on patients. Légaré and colleagues (Légaré 2011) reviewed published and unpublished literature regarding the effectiveness of patient and public engagement programs in clinical practice guidelines and described the barriers and facilitators to these programs. Barriers included recruiting challenges, lack of representation of certain patient and public groups, and participants' lack of familiarity with medical and scientific terminology. Facilitators included the provision of training and support. Similarly, Grant and colleagues (Grant 2018) conducted a rapid review and qualitative evidence synthesis to make inferences about how feasible online methods for patient engagement would be during clinical practice guideline development. The authors highlighted important considerations for patient and caregiver participation related to skills, time and resources. Notably, both of these reviews only considered patient and public involvement and did not consider the engagement of other stakeholder groups such as healthcare providers or policymakers, for example. Recently, Selva and colleagues (Selva 2017) reviewed guidance documents for developing clinical guidelines to assess how these documents address the

incorporation of patients' views. They identified the need for additional guidance in this area.

Cluzeau and colleagues (Cluzeau 2012; Kelson 2012; Kunz 2012) conducted a literature review and workshop to answer six questions related to stakeholder engagement in guideline development, including identification of the potential barriers and facilitators to integrating stakeholder involvement. They defined stakeholders as "all those who have a legitimate interest in a guideline. They include healthcare professionals, patients and caregivers, public and private funding bodies, managers, employers, and manufacturers". They noted that integrating different, sometimes competing, stakeholder perspectives can be challenging. Other barriers included issues with potential bias among stakeholder views, and the costly nature of the process. In contrast, they found that educating stakeholders and ensuring effective communication enabled effective stakeholder engagement.

In sum, there is a lack of systematic reviews that have synthesised barriers and facilitators to multi-stakeholder engagement in health guideline development, and our review aims to address this gap in the literature. The results of this review will be used to inform the development of guidance for multi-stakeholder engagement in guideline development and implementation. Specifically, the results of this review will be used by the MuSE Consortium to develop a Stakeholder Engagement Extension to the GIN-McMaster Checklist for guideline development. The findings of this review will assist organisations who develop healthcare, public health, and health policy guidelines to involve multiple stakeholders in the guideline development process to ensure the development of relevant, high quality, and transparent guidelines.

#### **4.4 OBJECTIVES**

The objective of this review is to identify and synthesise the existing evidence on barriers and facilitators to stakeholder engagement in health guideline development. We will address this objective through two research questions:

1. What are the barriers to multi-stakeholder engagement in health guideline development across the 18 topics of the GIN-McMaster checklist?
2. What are the facilitators to multi-stakeholder engagement in health guideline development

across the 18 topics of the GIN-McMaster checklist?

## **4.5 METHODS**

### **Reflexivity and positionality: Stakeholder engagement in the conduct of this review**

This review was developed and conducted by members of the MuSE Consortium, a group of individuals interested in advancing the methodology of stakeholder engagement in research and health care. The protocol for this review (Magwood 2022; See Appendix 8) was developed by a multi- stakeholder research team that engaged investigators, trainees, providers of care, policymakers, peer review editors and patient representatives. All members of the authorship team were provided with equal opportunities to be engaged in selection of studies, data extraction, critical appraisal, analysis and writing.

### **Criteria for including and excluding studies**

#### **Types of studies**

We included primary research studies which qualitatively reported on the barriers or facilitators to stakeholder engagement in health guideline development. This included qualitative or mixed method research studies using methods such as interviews, focus groups, or surveys to collect participant experiences; case studies of existing programmes; and process evaluation studies.

We included published guidelines that reported lessons learned only if these statements were derived from empirical qualitative methods and the details of these methods are available in a published report. Mixed method and intervention studies (randomised or non-randomised designs) were eligible only if they qualitatively report barriers and facilitators to engagement, independent of their quantitative findings.

We excluded non-empirical publications including commentaries and editorials. We excluded publications with incomplete data, including conference abstracts and protocols. Finally, we excluded existing evidence syntheses (e.g., systematic reviews, meta-ethnographies, scoping

reviews) as it would not be possible to extract the information relevant to our review, and reviewed their reference lists to identify any potentially eligible primary research articles.

### **Types of participants**

The population of interest is stakeholders in health guideline development. We define stakeholders as “any individual or group who is responsible for or affected by health- and healthcare-related decisions.” (Concannon 2012). Building on previous work, we have identified 10 types of stakeholders whose input can enhance the relevance and uptake of guidelines (Concannon 2012; Concannon 2019; Tugwell 2006). We included studies which reported on the barriers or facilitators to engagement of one or several of these stakeholder groups at any step of the guideline development process. For the purposes of this review, have grouped them as follows:

- Patients, caregivers, and patient advocates
- Public
- Providers of health care
- Payers/purchasers of health services
- Payers of research
- Policy makers
- Program managers
- Product makers
- Principal investigators and their research teams, and
- Peer-review editors/publishers

We recognise that within each of these categories, there is diversity and heterogeneity in types of stakeholders. For example, we recognise that patients and caregivers have differing characteristics and experiences, and their engagement brings diverse value to guideline development. The categories outlined above are for operational purposes and are based on prior research (Concannon 2012; Concannon 2019; Tugwell 2006). Studies which consider a single group of stakeholders or multiple groups of stakeholders were eligible for inclusion. If a study included multiple stakeholder groups, findings will be coded for each stakeholder group separately.

## **Phenomena of interest**

Eligible studies must have described stakeholder engagement in any of the topics of the GIN-McMaster Checklist for Guideline Development (Schunemann 2014). Eligible studies must have reported primary qualitative data in their results on barriers or facilitators to stakeholder engagement for at least one topic of the checklist:

1. Organization, budget, planning and training
2. Priority-setting
3. Guideline group membership
4. Establishing guideline group processes
5. Identifying target audience and topic selection
6. Consumer and stakeholder involvement
7. Conflict of interest considerations
8. Question generation
9. Considering importance of outcomes and interventions, values, preferences, and utilities
10. Deciding what evidence to include and searching for evidence
11. Summarizing evidence and considering additional information
12. Judging quality, strength or certainty of body of evidence
13. Developing recommendations and determining their strength
14. Wording of recommendations and of considerations about implementation, feasibility and equity
15. Reporting and peer review
16. Dissemination and implementation
17. Evaluation and use
18. Updating

## **Outcomes of interest**

The primary outcomes of interest were any barriers and facilitators to stakeholder engagement in health guideline development. Barriers were defined as any variable or condition that impedes stakeholder engagement in health guideline development. Facilitators were defined as any variable or condition that promotes stakeholder engagement in health guideline development. To be eligible, these outcomes could be reported as first or second order constructs related to

stakeholders' and study team perceptions and experiences of barriers and facilitators to their engagement in guideline development. First-order constructs refer to original narratives (e.g., participant quotes and observations) in the reviewed articles that relate to a particular topic. The interpretations of these narratives by the authors of the articles (e.g., themes, author explanations or recommendations) are considered second-order constructs, while the synthesis of the review team are third-order constructs (Figure 3). In this review, both first order and second order constructs were considered eligible data for inclusion, extraction and analysis and could be located in an articles' method, results, discussion or conclusion sections of the report.

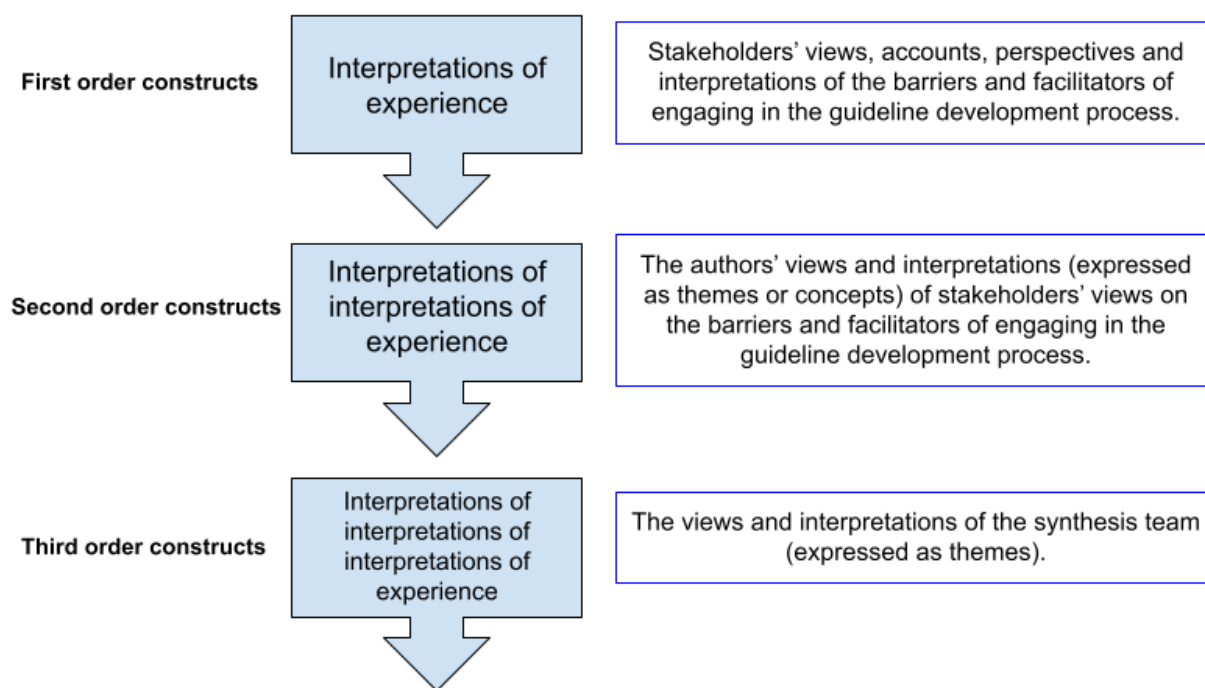


**Figure 3. First, second and third order-constructs of qualitative evidence**

## Setting

We did not place any restrictions on geographic settings. Studies focused on health guideline development from any country in the world were eligible for inclusion.

**Language**

No language restrictions were applied to our search, however all identified included studies were published in English.

**Publication date**

No publication date restrictions were applied.

**Search methods for identification of studies**

This review is part of a series of four reviews conducted by the MuSE working group on stakeholder engagement in guideline development. One comprehensive search strategy was developed and peer-reviewed in consultation with a medical librarian in accordance with PRESS guidelines (McGowan 2016). A second medical librarian reviewed the search strategy.

**Electronic databases**

The search combined index terms and keywords, combining concepts of stakeholder engagement (e.g., “patient participation”, “consumer involvement”) and guidelines (e.g., “clinical practice guideline”). Complete search strategies for all databases are available in Appendices 1-6.

We searched the following databases from database inception up to the 26th of September, 2022:

- MEDLINE (OVID) (Appendix 1)
- Cumulative Index to Nursing & Allied Health Literature (CINAHL; EBSCO) (Appendix 2)
- EMBASE (OVID) (Appendix 3)
- PsycInfo (OVID) (Appendix 4)
- Scopus (Appendix 5)
- Sociological Abstracts (Appendix 6)



## **Searching other resources**

We conducted an extensive grey literature search using the websites of agencies who actively engage stakeholders in research and/or guideline-producing agencies. We searched the following resources between June 24 and July 29, 2020:

- Agency for Healthcare Research and Quality's (AHRQ)
- Canadian Institutes of Health Research (CIHR) Strategy for Patient-Oriented Research (SPOR)
- National Institute for Health and care Research (NIHR)
- Be Part of Research
- Guidelines International Network (G-I-N)
- INVOLVE
- National Institute for Health and Care Excellence (NICE)
- Patient-Centered Outcomes Research Institute (PCORI)
- American Academy of Paediatrics
- Australia's National Health Medical Research Council (NHMRC)
- World Health Organization (WHO), including Latin American and Caribbean Health Sciences Literature (LILACS)

We solicited suggestions for additional grey literature sources from the members of the MuSE Consortium. Backward and forward citation tracking was performed on included articles to identify further eligible studies. We reviewed the reference lists of relevant reviews to identify eligible primary studies for inclusion.

## **Selection of studies**

All identified citations from electronic databases were imported into Covidence software for screening and selection. Documents identified through our grey literature search were managed and screened using an Excel spreadsheet. A two-part study selection process was used for all identified citations: (1) a title and abstract review and (2) full-text review. The selection process

was piloted by senior review authors (O.M., J.P., J.K., L.L., A.R., P.A.) and a moderate to very good level of agreement (Cohen's Kappa range 0.43-0.87) was achieved before all studies were assessed for eligibility, independently and in duplicate. We resolved any disagreements through discussion during weekly meetings for all planned reviews. We produced a PRISMA flow diagram which reports the number of studies included and excluded at each stage. Reasons for exclusion have been provided for all studies assessed at full-text.

### **Details of study coding categories**

Data were extracted by two review authors independently and in duplicate according to a standardised data extraction form. Disagreements were discussed and resolved by a third reviewer. The data extraction form was pilot tested by two review authors and coding categories were clarified iteratively, as needed. Data were extracted on basic study characteristics and methods including:

- Study aim/objective
- Study setting (i.e. location where the guideline development took place, and geographical location)
- Type of guideline (e.g., clinical practice, public health, or health system guideline, specific condition(s) addressed)
- Population and participant characteristics, including categorisation according to stakeholder group and equity-relevant characteristics according to PROGRESS-Plus (O'Neill 2014). PROGRESS-Plus is an acronym referring to Place of Residence, Race/ethnicity, Occupation, Gender, Religion/culture, Education, Socio-economic status, Social capital/networks) and other important factors which impact on health equity (e.g. disability, sexual orientation, age).
- Recruitment methods
- Data collection methods
- Analysis methods

In addition, we extracted information on study:

- Limitations

- Recommendations
- Conclusions
- Funding
- Conflict of Interest

For qualitative outcomes, we extracted data using codes adopted from the Theoretical Domains Framework (TDF) (Atkins 2017). Implementing new or changing existing stakeholder engagement practices in organisations who develop guidelines requires a change in individual and collective behaviour. Changing behaviour requires an understanding of the influences on behaviour in the context in which they occur. Developed through a multidisciplinary consensus approach and subsequent validation, TDF consolidates overlapping behavioural theories into 14 domains encompassing 84 theoretical constructs, and provides a theoretical lens through which to view the cognitive, affective, social and environmental influences on behaviour and implementation. We utilised the 14 overarching domains as deductive categories for extraction [See Table 3]. We introduced an “other” category to capture data which did not fit under any TDF domain.

**Table 3.** Data extraction categories from the Theoretical Domains Framework (Atkins 2017).

| <b>TDF Domain</b>                     | <b>Definition</b>                                                                                                             | <b>Examples</b>                                                                                                                                                              |
|---------------------------------------|-------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Knowledge                             | An awareness of the existence of something                                                                                    | Knowledge (including knowledge of condition /scientific rationale); Procedural knowledge; Knowledge of task environment                                                      |
| Skills                                | An ability or proficiency acquired through practice                                                                           | Skills; Skills development; Competence; Ability; Interpersonal skills; Practice; Skill assessment                                                                            |
| Social/Professional Role and Identity | A coherent set of behaviours and displayed personal qualities of an individual in a social or work setting                    | Professional identity; Professional role; Social identity; Identity; Professional boundaries; Professional confidence; Group identity; Leadership; Organisational commitment |
| Beliefs about Capabilities            | Acceptance of the truth, reality, or validity about an ability, talent, or facility that a person can put to constructive use | Self-confidence; Perceived competence; Self-efficacy; Perceived behavioural                                                                                                  |

|                                          |                                                                                                                                                                                         |                                                                                                                                                                     |
|------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|                                          |                                                                                                                                                                                         | control; Beliefs; Self-esteem; Empowerment; Professional confidence                                                                                                 |
| Optimism                                 | The confidence that things will happen for the best or that desired goals will be attained                                                                                              | Optimism; Pessimism; Unrealistic optimism; Identity                                                                                                                 |
| Beliefs about Consequences               | Acceptance of the truth, reality, or validity about outcomes of a behaviour in a given situation                                                                                        | Beliefs; Outcome expectancies; Characteristics of outcome expectancies; Anticipated regret; Consequents                                                             |
| Reinforcement                            | Increasing the probability of a response by arranging a dependent relationship, or contingency, between the response and a given stimulus                                               | Rewards (proximal / distal, valued / not valued, probable / improbable); Incentives; Punishment; Consequents; Reinforcement; Contingencies; Sanctions               |
| Intentions                               | A conscious decision to perform a behaviour or a resolve to act in a certain way                                                                                                        | Stability of intentions; Stages of change model; Transtheoretical model and stages of change                                                                        |
| Goals                                    | Mental representations of outcomes or end states that an individual wants to achieve                                                                                                    | Goals (distal / proximal); Goal priority; Goal / target setting; Goals (autonomous / controlled); Action planning; Implementation intention                         |
| Memory, Attention and Decision Processes | The ability to retain information, focus selectively on aspects of the environment and choose between two or more alternatives                                                          | Memory; Attention; Attention control; Decision making; Cognitive overload / tiredness                                                                               |
| Environmental Context and Resources      | Any circumstance of a person's situation or environment that discourages or encourages the development of skills and abilities, independence, social competence, and adaptive behaviour | Environmental stressors; Resources / material resources; Organisational culture /climate; Salient events / critical incidents; Person x environment interaction     |
| Social influences                        | Those interpersonal processes that can cause individuals to change their thoughts, feelings, or behaviours                                                                              | Social pressure; Social norms; Group conformity; Social comparisons; Group norms; Social support; Power; Intergroup conflict; Alienation; Group identity; Modelling |
| Emotion                                  | A complex reaction pattern, involving experiential, behavioural, and physiological elements, by which the individual attempts to deal with a personally significant matter or event     | Fear; Anxiety; Affect; Stress; Depression; Positive / negative affect<br>Burn-out                                                                                   |

|                        |                                                                                 |                                                  |
|------------------------|---------------------------------------------------------------------------------|--------------------------------------------------|
| Behavioural Regulation | Anything aimed at managing or changing objectively observed or measured actions | Self-monitoring; Breaking habit; Action planning |
|------------------------|---------------------------------------------------------------------------------|--------------------------------------------------|

We extracted stakeholders' (first-order constructs) and author (second-order constructs) perceptions and experiences regarding the barriers and facilitators to engagement in health guideline development from the results and discussion sections, including participant quotations from interviews or focus groups, excerpts or quotations from documentary analysis, narrative descriptive summaries, author hypotheses, explanations and recommendations, themes and sub-themes. Discrepancies in the data extraction process were resolved through discussion.

## Analysis

Our review included studies employing a qualitative approach to inquiry. We used the 'best fit' framework method as a systematic and flexible approach to analysing the qualitative data (Booth 2015; Carroll 2013; Gale 2013). Framework-based synthesis, using the 'best fit' method, is a highly pragmatic and useful strategy for analysing a range of questions (Dixon-Woods 2011) and is supported by guideline development literature (Flemming 2019). Framework analysis is a five-stage process that includes familiarisation with the data, identifying a thematic framework, indexing (applying the framework), charting and mapping, and interpretation (Spencer 1994). Data were analysed deductively, using the TDF domains to generate a framework of preliminary themes. Whilst the domains are purposefully designed to be broad groupings of the possible factors to influence behaviour, the intent is to explore the important domains in further detail. Two review authors (O.M. and A. Raymond) examined the data that were extracted under each TDF domain. The two authors identified and came to agreement on recurring concepts to develop the initial themes. Disagreements were resolved through discussion. When necessary, the authors returned to the full texts of the included studies to verify the perspective and context of the extracted data. After developing the initial thematic findings, all themes were circulated to the authorship team for comment. In a series of iterative drafts, the themes were refined and expanded upon. An illustrative quote was selected for each theme and the authorship team decided to organize themes using tabular and narrative approaches.

All identified key findings were mapped to the 18 topics of guideline development process

outlined by the GIN-McMaster checklist (Schunemann 2014). We present this information as a narrative and series of tables, indicating the barriers and facilitators that exist for each stakeholder group and for each step of guideline development. The findings of this mapping exercise will be combined with the results of the other reviews in this series (Khabisa 2022; Lytvyn (in press); Petkovic 2022) to develop international guidance on stakeholder engagement in guideline development.

### **Dealing with missing data**

Missing data is an inherent component of the qualitative research process. Since meanings and interpretations are the outputs of qualitative studies, data are always missing because there are some questions that respondents do not want to answer, others they circumvent, and aspects of their experiences which may not be reported (Singh 2013). In such situations, the silence on these issues in itself becomes powerful data. Our findings have been mapped against the GIN-McMaster Checklist and we report which steps of the checklist do have supporting qualitative evidence and those which do not, thereby highlighting gaps in evidence and opportunities for new qualitative inquiry. We did not contact authors of included studies to inquire about missing data.

### **Critical appraisal**

Two review authors assessed the methodological rigour of all included studies independently and in duplicate. We assessed the quality of qualitative studies and the qualitative components of mixed method studies using the Critical Appraisal Skills Programme (CASP) qualitative appraisal research tool (Singh 2013). Discrepancies in the critical appraisal process were resolved by a third reviewer.

### **Confidence in findings**

We conducted GRADE-CERQual assessments to determine the confidence in the qualitative findings. Assessments were conducted by one review author (P.C.) and reviewed by a second

author (O.M.). GRADE-CERQual is an assessment of the extent to which the review finding is a reasonable representation of the phenomenon of interest, and results in four levels based on the findings relevance, methodological limitations, coherence, and adequacy of the data (Lewin 2018):

- *High confidence in the evidence*: It is highly likely that the review finding is a reasonable representation of the phenomenon of interest.
- *Moderate confidence in the evidence*: It is likely that the review finding is a reasonable representation of the phenomenon of interest.
- *Low confidence in the evidence*: It is possible that the review finding is a reasonable representation of the phenomenon of interest
- *Very low confidence in the evidence*: It is not clear whether the review finding is a reasonable representation of the phenomenon of interest.

GRADE-CERQual assessments for each finding are available in Appendix 7

## 4.6 RESULTS

### Results of the search

Our systematic search identified 55,364 citations. After removal of duplicates, we screened 31,505 articles by title and abstract. Of these, 731 were retrieved for full-text review.

Additionally, our citation analysis and grey literature search identified 83 reports which warranted full-text review. After review of all articles against a priori inclusion criteria, we included a total of 34 studies published in 40 reports. Thirty-eight reports were identified through database searching and 2 reports were identified via other methods (Figure 4).

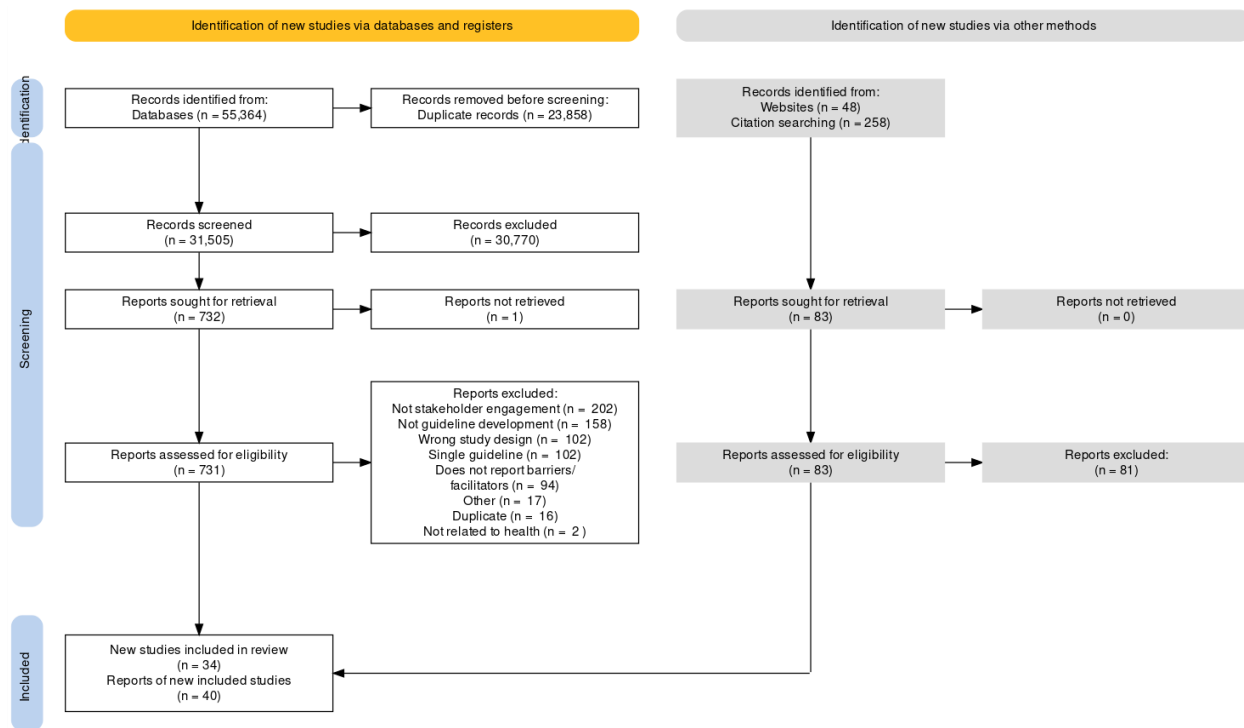


**Figure 4. PRISMA Flow Diagram**

## Included studies

We included 34 studies (40 publications) which qualitatively reported barriers or facilitators to stakeholder engagement in any topic of the guideline development process. The characteristics of these studies are described in Table 4. The majority of included studies are observational mixed method or qualitative designs, employing methods such as surveys, interviews, focus groups and participant observations. We included one single-blind crossover pragmatic study (Lamontagne 2021). The included studies captured perspectives from at least 3400 participants. The number of participants was not reported in several studies. Most participants were recruited through convenience or purposeful sampling, drawing on stakeholders from established programs or networks. Study samples are summarised below according to PROGRESS+ characteristics. No information was reported on religion, socio-economic status, social capital, or other dimensions associated with disadvantage.

*Place of residence:* All included studies except for one (Kredo 2018) were conducted in high-



income countries: Canada (n=7), UK (n=7), USA (n=5), Netherlands (n=4), Finland (n=2), Europe more broadly (n=3) and Australia (n=1). Four studies collected data internationally, but were managed by a project team based out of Canada (n=3) or Australia (n=1). Kredo 2018 was conducted in South Africa, an upper-middle-income country with significant disparities between residents. In this study, one participant explained how South African provinces have vastly different resources, cultures and infrastructure, and that a failure to accommodate these diversities could have dire consequences (Kredo 2018). Notably, no evidence was identified on the barriers or facilitators to engaging stakeholders originating from low-income countries.

One study recruited patient representatives with traumatic brain injuries from a community-based association, and highlighted that this association was chosen because it encompassed rural and urban areas (Lamontagne 2021). Of the 16 patients included in the study, 9 (56.3%) lived alone and 7 (43.4%) lived with close relatives. Only one other study reported on the living situation of their participants; Lanza 2000 took place within the psychiatric ward of a veterans hospital and recruited in-patients who resided on the ward.

*Race/ethnicity/ancestry/language/culture:* Six studies reported on the race of their participants: 4/6 reported that the majority of their sample identified as white (range 87.5-100%) (Armstrong 2019; Armstrong 2020; Harding 2010; Lanza 2000). One study reported the majority of their sample was African-American (67% n=10) (Armstrong 2017), and in another study one consumer self-identified as belonging to a minority ethnic group (Synnot 2022). In most studies, the ability to communicate in English was a requirement for study participation. One study was conducted among French-speaking individuals living in a majority Francophone context (Lamontagne 2021).

*Occupation:* Participants of the included studies covered 9/10 MuSE stakeholder groups, including: i) Patients, caregivers, and patient advocates, ii) Public, iii) Providers of healthcare, iv) Payers of health services, v) program managers, vi) policymakers, vii) Product makers, viii) peer review journal editors and ix) Principal investigators and their research teams. No studies included payers of research. One study reported a median of 9 years of professional experience among stakeholders (Median: 9; range: 1-26) (Den Breejen 2014), whereas another

study reported that employment status among patient representatives varied (Köpke 2019).

*Gender and sex:* Thirteen studies reported on the gender or sex of their participants. In 7/13 studies, the samples were described as majority female or women (range 56-87%) (Armstrong 2017; Armstrong 2019; Biggane 2022; Brouwers 2017; Köpke 2019; Lindsay 2020; Synnot 2022). Three studies reported their samples as the majority being males (Armstrong 2020; Lamontagne 2021; Lanza 2000). In three additional studies, the samples had equal representation from males and females (Chartres 2020; Den Breejen 2014; Harding 2010). Notably, no studies reported on diverse gender identities.

*Religion:* Not reported

*Education:* While infrequently reported, three studies described the level of education of their participants. In their study with cancer patients, cancer survivors, and their families/caregivers, Brouwers 2017 described that 85% (n=34) had post-secondary degrees or diplomas. Similarly, Köpke 2019 reported that 57% of patients and 64% of caregivers had tertiary education. In contrast, Lanza 2000 reported that most in-patient residents had either high school or college education levels.

*Socio-economic status:* Not reported.

*Social capital:* Not reported.

+ *Age:* One study explicitly aimed to develop a tool for guideline developers to take children's perspectives into account and facilitate children's participation in the process of guideline development (Schalkers 2017). However, no children were included as participants in the study and data was collected from professionals involved in guideline development. An additional 8 studies reported on the age of their participants, varying in age from mid 20s to late 70s (Biggane 2022). In 4 studies, the majority of the sample was described as "middle aged" (40-60 yrs) (Den Breejen 2014; Köpke 2019; Lamontagne 2021; Lanza 2000), and in 3 studies the majority of the sample was described as "older" (60+ years) (Armstrong 2017; Brouwers 2017; Synnot 2022).

+ *Disability*: Two studies aimed to engage people living with disability. In their study engaging patients with dementia, Armstrong 2020 recruited four patient representatives. One of these was an individual with mild cognitive impairment/early Alzheimer's disease, who was able to fully participate alongside their spouse. In another study, Lamontagne recruited patients with traumatic brain injury, and 10/16 (62.5%) of patient representatives had a severe traumatic brain injury (Lamontagne 2021).

No other dimensions of "PLUS" (personal characteristics associated with disadvantage) were reported in the included studies.

**Table 4: Characteristics of Included Studies**

| Study ID                                                          | Study Design  | Aim                                                                                                                                                                                                                         | Setting                                                                    | Participants                                                                                                                                                              | Data Collection                                                                                                                                                                                                                                                                                                                                                           | Data Analysis                                                                                                                                                                                                                                   |
|-------------------------------------------------------------------|---------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Armstrong 2017                                                    | Qualitative   | To investigate the perspectives of potential patient/consumer guideline representatives on topics pertaining to engagement including guideline development group composition and barriers to and facilitators of engagement | University of Maryland and University of Florida, United States of America | Participants were members of a program designed to link community members to academic research, and had no prior experience participating in guideline development (n=15) | Focus groups, facilitated using a topic guide. Focus groups lasted up to 1 hour. All groups were audio recorded and transcribed verbatim.                                                                                                                                                                                                                                 | Qualitative analysis was performed according to a qualitative descriptive approach and was used to identify, define, and organize themes. Results were compared to an evidence-based framework for patient and consumer engagement in research. |
| Armstrong 2019<br><br>Companions: Khodyakov 2017, 2019 Grant 2021 | Mixed-methods | To describe participant experiences with a novel online, scalable approach for patient and caregiver engagement in CPG development.                                                                                         | Online ExpertLens™ system, RAND Corporation, USA                           | Participants were patients with Duchenne Muscular Dystrophy (DMD) (n=24) and their caregivers (n=71).<br><br><b>Khodyakov 2017:</b><br>Study 1: (n=43)<br>Study 2: (n=50) | All participants participated in a satisfaction survey. Further, 5 patients and 20 caregivers participated in semi-structured telephone interviews.<br><br><b>Khodyakov 2017:</b> OMD using RAND's ExpertLens involving Likert Scales and asynchronous/anonymous discussion, Face to face meeting for study 3, Optional Survey about experiences with the online process. | Descriptive analyses of responses to satisfaction questions. To qualitatively analyze the interview data, a codebook was developed and                                                                                                          |

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|--|--|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|  |  | <p><b>Khodyakov 2017:</b><br/>To explore the acceptability of the Online Modified Delphi (OMD) panel approach from the patients perspective.</p> <p><b>Khodyakov 2019:</b><br/>To present the RAND/PPMD (Patient-Centeredness Method) a novel online approach to engaging patients and their representatives in CPG development.</p> <p><b>Grant 2021:</b> To propose ways that guideline development groups can use online modified-Delphi processes to engage a large panel of patients in a manner compatible with best-practices at each stage of CPG</p> |  | <p>Study 3: (n=28) All studies involved physicians, allied health professionals, researchers, and patients. Study 3 also included clinic managers/administrators.</p> | <p><b>Khodyakov 2019:</b> The RPM requires iterative data collection. Patients and caregivers provide their input by generating ideas, rating and commenting on draft recommendations, discussing preliminary rating results, and revising their responses based on the panel's preliminary ratings and group discussions.</p> | <p>used an inductive thematic approach to qualitative data analysis.</p> <p><b>Khodyakov 2017:</b> 7-Point Likert scale were used to gauge acceptability of the OMD.</p> <p><b>Khodyakov 2019:</b> Our team evaluated participation experiences of the individuals with DMD and caregivers who helped us test the RPM.</p> |
|--|--|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|

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|                |                                       | development and consistent with frameworks for continuous patient engagement across the entire CPG development process. |                                                                                                                                     |                                                                                                                                                                                                                                                                                  |                                                                                                                                                                |                                                                                                                                                                                                                    |
| Armstrong 2020 | Qualitative (instrumental case study) | To assess the impact of participation and consultation strategies on guideline question development                     | American Academy of Neurology (AAN) guideline on amyloid positron emission tomography (PET) imaging in patients with dementia, USA. | N= 15. A patient, two caregivers and a dementia advocate participated in the guideline development group alongside 3 content experts, 3 guideline subcommittee members with content expertise, 4 guideline subcommittee members without content expertise, and 2 methodologists. | Public comment responses and guideline development group meeting transcripts.                                                                                  | Analysed descriptively. Transcript quotes were compared to the conceptual model of PPI in guideline development. The effects of participation and consultation strategies within the guideline case were compared. |
| Atkins 2013    | Qualitative                           | To investigate how evidence is translated into recommendations from the perspective of                                  | National Institute for Health and Care Excellence (NICE),                                                                           | Participants were members of the NICE advisory groups (acute physical health, mental health, and                                                                                                                                                                                 | Face-to-face or telephone interviews, depending on preference, were conducted in a location convenient to the participant. Interviews lasted 30 to 45 minutes. | A thematic analysis was conducted to identify recurrent themes in the data. This was                                                                                                                               |

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|                  |                          | NICE guidance groups                                                                                                                                                                                                          | United Kingdom                                                                                                           | public health) (n=39). This included 3 Chairs, 16 healthcare professionals, 9 technical team members (4 health economists, 2 project managers, 1 systematic reviewer, 1 research fellow, 1 information scientist), 7 service users, 2 professional members, and 2 service providers. |                                                                                                                                                        | followed by a content analysis of the number of utterances to provide quantitative data to contextualise identified themes.                                                                                                                     |
| Bertelman n 2020 | Qualitative (case study) | To measure stakeholder participation in the external review of the 2016 European acne guideline and assess the extent to which comments submitted by pharmaceutical companies suggested changes favouring their own products. | During the external review of the 2016 update of the European evidence-based guideline for the treatment of acne, Europe | n=10. Pharmaceutical companies (5/5 invited took part); professional association members (2/222 took part); journal reviewers (2); and visitors to the website (1).                                                                                                                  | Retrospective, partially blinded review and qualitative content analysis of all comments (n=74) submitted during the external review of the guideline. | Comments were coded deductively from the literature and inductively from a pilot coding of 10 comments. A user manual was developed to provide definitions and examples for each item. After two investigators had independently extracted data |

|              |                                                    |                                                                                                                                                                                                                                             |                                                               |                                                                                                                                                                                                                                                                                                                                      |                                                                                                                                                                                                                       |                                                                                                                                                                                                               |
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|              |                                                    |                                                                                                                                                                                                                                             |                                                               |                                                                                                                                                                                                                                                                                                                                      |                                                                                                                                                                                                                       | from all comments, discrepancies were identified by a third investigator.                                                                                                                                     |
| Biggane 2022 | Qualitative (Ethnographic Methodological Approach) | To explore how the input of Patient and Public Involvement (PPI) contributors at the National Institute of Health and Care Excellence (NICE) is integrated into guideline development, particularly in relation to health outcome selection | NICE (UK)                                                     | Participants were involved in NICE guideline development processes and were health professionals, care providers and lay members (patients, carers, service users, and patient representatives). Interviews were conducted with lay members (n=14), health professionals (n=2), and committee chairs (n=2). Female (n=10) Male (n=8) | In-situ observations of five different guideline development processes including clinical guideline meetings, scoping workshops, and lay member training sessions (over 230 Hr) In-depth semi-structured interviews   | Iterative and thematic analysis from a pragmatic theoretical standpoint. Data was coded and similar codes were grouped together to identify recurring patterns and organize these into themes and categories. |
| Boivin 2010  | Qualitative                                        | To develop an international practice and research agenda on patient and public                                                                                                                                                              | International workshop organised by GIN PUBLIC at the 5th GIN | A total of 56 individuals participated in the workshop. This included 35 guideline                                                                                                                                                                                                                                                   | Structured and facilitated discussion during a 3-hour workshop. An oral summary of discussions was fed back to all participants at the end of the workshop, and facilitators produced a written report of discussions | Distributed the original data to G-I-N PUBLIC Steering Committee members and                                                                                                                                  |



|               |               |                                                                                                                                                                                                                                                                           |                                                                                                                                                                                      |                                                                                                                                                          |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                                  |
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|               |               | involvement in CPG                                                                                                                                                                                                                                                        | conference in Helsinki (Finland), in November 2008                                                                                                                                   | developers, 16 researchers and 5 patient/public representatives. Attendees were from 14 different countries                                              |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                           | solicited written comments to validate findings, before discussing recommendations with them in two teleconferences.                                                                                                                                                                                                                                             |
| Brouwers 2017 | Mixed-methods | To obtain information from cancer patients, cancer survivors, and their families/caregivers about their awareness of PGs, attitudes towards PGs, interest in participating in PG development, potential barriers and facilitators to participation, and information needs | Workshops delivered in Ontario, Canada. Surveys given at Juravinski Hospital and Cancer Centre as well as at two local cancer support programs. Setting of interviews not described. | Forty-one people participated in the study. Twelve individuals attended workshops, 21 completed surveys, and eight participated in telephone interviews. | <p>Workshops: Facilitated discussion centering participants' awareness of, and attitudes towards, PGs, their interest in participating in PG development, potential barriers and facilitators to their participation, and their information needs.</p> <p>Survey: The survey was anonymous and was available for completion from February to April 2015.</p> <p>Interviews: Guided by a structured interview form that examined current processes for patients and caregivers in guideline activities</p> | <p>Workshops: Sessions were audio-recorded, transcribed verbatim and participant codes were used to ensure confidentiality of respondents. Transcripts were independently reviewed by two project members, who conducted inductive and deductive thematic analyses of the data.</p> <p>Survey: Quantitative survey data were analyzed using Microsoft Excel.</p> |

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|               |               |                                                                                                                                                                                |                                      |                                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                                  | Interviews: All conversations were audio-recorded. Interview summary forms were completed after the interview and sent to the interviewees for verification and revision. The forms were then independently reviewed by two project members, who conducted a thematic analysis of the data. |
| Brouwers 2018 | Mixed-methods | To test the feasibility and efficacy of two patient engagement models by Cancer Care Ontario's cancer guideline development program, the Program in Evidence-Based Care (PEBC) | Cancer Care Ontario, Toronto, Canada | Participants are Cancer Care Ontario's Patient and Family Advisors. These members are cancer patients, family members, and caregivers (n=9). Also included Health Research Methodologists (HRMs; n=8) and | A modified version of the Public and Patient Engagement Evaluation Tool (a questionnaire) was used to gather feedback from the patient participants about their level of satisfaction with the process and outcome of each model. An additional survey was developed by the study team to gather similar data from participating HRMs and working group members. | No methods of analysis reported.                                                                                                                                                                                                                                                            |

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|               |                                            |                                                                                                                                                                                                                                                                                            |                                               | working group members (n = not reported).                                                                                                                         |                                                                                                                                                                                                               |                                                                     |
| Carter 1995   | Quantitative descriptive (cross-sectional) | To survey representatives of guideline organisations on their activities and attitudes toward organisational roles, setting of priorities, implementation of guidelines, evaluation, and terms of reference                                                                                | Canada                                        | Organisations identified as being actively involved in or considering active involvement in the CPG field at the national or provincial/territorial level (n=55). | Questionnaires; Organizations were asked to mail completed questionnaires to the CMA. Nonresponding organisations were telephoned close to the response deadline and reminded that the questionnaire was due. | Questionnaire responses were analysed in a descriptive manner only. |
| Chalmers 2017 | Descriptive (case report)                  | (1) To understand the different ways in which patients can contribute to clinical guidelines, research projects and educational activities. (2) To understand the barriers and potential solutions to these barriers from a physician's perspective, in order to ensure meaningful patient | Europe; 150 centres in more than 40 countries | Patients living with Bronchiectasis and their carers (n=711)                                                                                                      | An online patient survey co-ordinated by European Lung Foundation.                                                                                                                                            | Not reported                                                        |

|               |             |                                                                                                                                                                                                                                                                                                                                                        |                                                                 |                                                                                                        |                                                                                        |                                                                                                                                                                                                  |
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|               |             | involvement in clinical projects.<br>(3) To understand the barriers and potential solutions from a patient's perspective, in order to meaningfully involve patients in clinical projects.                                                                                                                                                              |                                                                 |                                                                                                        |                                                                                        |                                                                                                                                                                                                  |
| Chartres 2020 | Qualitative | To empirically examine the perspectives and experiences of the key participants involved in developing public health guidelines for the Australian National Health and Medical Research Council (NHMRC). Specifically, to gain a better understanding of the social processes involved in development of health-related guidelines, e.g. how guideline | National Health and Medical Research Council (NHMRC), Australia | N=20 (10 male and 10 female). Sixteen working committee members and four evidence review group members | In-depth semi-structured interviews, digitally recorded and professionally transcribed | Field notes written after each interview; transcripts analysed as received with line-by-line coding; case-based and conceptual memos written throughout the study and discussed at team meetings |

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|                         |               | group members interact and communicate, and how evidence is considered.                                                                                                                                                                                                                                                                                                                                                      |                                                                                         |                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                                                       |                                                                                                                                                                               |
| Daraz 2019 & Murad 2019 | Mixed-methods | <p>1. To develop clinical practice recommendations addressing management areas of highest importance to patients.</p> <p>2. To develop a training curriculum that can aid in a meaningful patient engagement during clinical practice guideline development and evaluate its implementation.</p> <p>3. To describe challenges faced by the guideline panels and specific innovations and solutions used to address them.</p> | American Society of Hematology , Washington , DC and Mayo Clinic Methodology group, USA | 62 experts (hematologists, SCD specialists, pain specialists, emergency medicine specialists, psychologists, neurologists, neuroradiologists, researchers in cognitive science education and psychology, cardiologists, pulmonologists and nephrologists, pathologists/transfusion specialists and transplant specialists). 10 patient representatives | Panel discussions where the members rated the importance of outcomes on a scale of 1 to 9. Training patients for meaningful engagement, and a post-curriculum survey. | Descriptive statistics and quotes of open ended questions were used to understand the perceptions. However, no inferential statistics or qualitative data analyses were used. |

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| Den Breejen 2014 | Mixed-methods | To assess the feasibility of a patient-centered network approach in a detailed evaluation of the process professionals' experiences, and the time required for guideline development                       | Dutch program of Knowledge Quality and Curative Care, Radboud University Nijmegen, The Netherlands | Thirty-two participants were involved (52% male), Age: (15% 26-35, 33% 36-45, 41% 46-55, 11% 55+), years of professional experience (Median 9; 1-26), 59% with experience in guideline development | Mixed-method evaluation including examination of secondary resources (such as project descriptions), interviews with key figures (n=8), two focus groups, and a written questionnaire survey among all participants                                                                                                                                                                                  | Descriptive statistics (frequencies, medians, and ranges) on participant background characteristics and to analyze the time data. Qualitatively analyzed the transcripts of the interviews and the free text responses from the second part of the questionnaire using an author-developed framework |
| Duff 1996        | Qualitative   | (1) To identify means by which patients and users of services can most effectively, and to their satisfaction, be involved in the development of national clinical guidelines. (2) To identify key factors | Seminar delivered by Dynamic Quality Improvement Programme, Royal College of Nursing, London, UK   | Seminar participants were patient representatives, health professionals, researchers and actual patients. No other details provided.(n= not reported)                                              | A national (UK) seminar was planned and coordinated to look at the involvement of patients and service users in the development and use of clinical guidelines. To promote debate the seminar participants were divided into three groups and worked with a facilitator with the aim of enabling all participants to contribute. Groups were allocated questions on which to focus their discussion. | All presentations and feedback sessions were audio tape recorded and notes taken. In the days following the seminar, seminar notes were collated and key issues, themes                                                                                                                              |

|                     |             |                                                                                                                                                                                                                                                                                                       |                                         |                                                                                                                                                                                                                                             |                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                    |
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|                     |             | influencing the effective involvement of patients and users of services in the development of national clinical guidelines. (3) To gain the support of participants for a research study to investigate the involvement of patients/service users in the development of national clinical guidelines. |                                         |                                                                                                                                                                                                                                             |                                                                                                                                                                                                                                                                     | and suggestions for guidance for collaboration identified by steering group members and suggestions for their resolution proposed.                                                                                 |
| Hameen-Antilla 2016 | Qualitative | To 1) discover ways to involve patients in HTA and CPG processes, 2) describe challenges, and 3) find ways of informing patients about HTAs and CPGs in Finland.                                                                                                                                      | One-day invitational seminar in Finland | Twenty-nine representatives from 19 patient organizations and 6 other organizations participated in the seminar. All participants were from Finland and included patient organization representatives, patients, health care professionals, | Focus group discussions (FGDs). The FGDs lasted for approximately 60 min and were tape-recorded and transcribed verbatim for the purpose of the analyses. Each FGD had two facilitators who followed an interview guide covering key themes with probing questions. | Data analysis was conducted by two researchers who acted as facilitators in the FGDs. They first thematically analyzed the data independently and interpretations were then compared. Discrepancies were discussed |

|                        |             |                                                                                                                                                                                                                                                                                                                                               |                                                       |                                                                                                                                                      |                                                                                                                                                                                                                                                                                                                    |                                                                                                                                                                                                                                                                                                                                  |
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|                        |             |                                                                                                                                                                                                                                                                                                                                               |                                                       | consumers and health authorities.                                                                                                                    |                                                                                                                                                                                                                                                                                                                    | and an agreement sought                                                                                                                                                                                                                                                                                                          |
| Harding 2010 & 2011    | Qualitative | To explore service users' experiences of their involvement in developing NICE guidelines.                                                                                                                                                                                                                                                     | United Kingdom                                        | Ten service users. All self-identified as being of white British ethnicity; five were male                                                           | A semi-structured interview schedule was developed that explored perceptions of data generated by scientific methods and experiential “phenomenological data”, relationships between group members and the guideline development process. Following a prescribed social constructivist grounded theory methodology | Data were continually analyzed using the constant comparison method as prescribed in grounded theory.                                                                                                                                                                                                                            |
| Illot 2006 & Rapu 2005 | Qualitative | To quantify stakeholder involvement from an organizational and individual perspective; to explore perceptions about the influence and impact of professional stakeholders; and to make recommendations about more effective and efficient ways in which professional stakeholders could contribute to the NICE guideline development process. | National Institute for Clinical Excellence (NICE), UK | Forty-six participants from professional bodies including healthcare providers and coordinators responsible for coordinating stakeholder engagement. | Two structured questionnaires were developed to quantify the involvement of professional body coordinators and expert members in the guideline development process. Semi-structured interviews explored the purpose, costs and benefits of stakeholding                                                            | Numerical data were evaluated with descriptive analysis of frequencies. The open item replies were transcribed verbatim, emerging themes identified using a constant comparative approach. For interviews, themes within and between each question, and links with the open item replies on the questionnaires, were identified. |



|             |             |                                                                                                                                                                                                                                                                           |                                                                                        |                                                                                                                                                                                                                                                         |                                                                                                                                                       |                                                                                                                                                                                                                |
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| Jarret 2004 | Qualitative | To explore the experiences of patient/carer members (PCMs) and chairs of Guideline Development Groups, to report on their experiences of patient/carer involvement on the groups, identify good practice, highlight problems, and to improve the process for future GDGs. | National Institute for Clinical Excellence (NICE), UK                                  | The study included 36 PCMs and 19 chairs. Among PCMs, 22 had a personal experience of the condition or topic under review, 5 were carers, 20 were employees of patient/carer organizations, 1 was a staff member from the Patient Involvement Unit      | The PCMs and chairs were approached by the Patient Involvement Unit (PIU) and invited to take part in a telephone interview or an in-person interview | Not reported                                                                                                                                                                                                   |
| Kim 2021    | Qualitative | To explore how developers generate guidelines formed by patient preferences                                                                                                                                                                                               | International (Canada, US, Australia, Ireland, Scotland, England and the Netherlands ) | Participants (n=50) had experience in guideline development. They were clinicians (n=16), patients (n=17) or managers (n=17). They were different types of guideline developers such as: government (n= 19), academic organization (n=4), international | Qualitative interviews by telephone                                                                                                                   | Themes were identified inductively using constant comparison, a preliminary codebook of themes and exemplar quotes was developed after the first two interviews and subsequent interviews were coded to expand |

|            |               |                                                                                                                                                                                                             |                                                                                                                                                                |                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                         |                                                                                                                                                                                                   |
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|            |               |                                                                                                                                                                                                             |                                                                                                                                                                | collaboration (n=3), professional society (n=16), or disease charity/foundation (n=8).                                                                    |                                                                                                                                                                                                                                                                                                                         | or merge themes with review from a second researcher.                                                                                                                                             |
| Kopke 2019 | Mixed-methods | To engage multiple sclerosis (MS) patients and caregivers in the definition of the key questions to be answered in the European Academy of Neurology guideline on palliative care of people with severe MS. | Europe<br><br>The focus groups were held in Milan (Foundation C. Besta), Berg (Marianne-Strauß-Klinik) and Hamburg (in a local patient self-help organization) | 934 participants from seven countries. Participants were MS patients (n=751, 74% women) and caregivers (n=183, 72% women).                                | Survey and focus group discussions                                                                                                                                                                                                                                                                                      | Qualitative coding of open sections. Categorical variables were summarized as numbers and percentages, continuous variables were summarized as means and SD or medians with interquartile ranges. |
| Kredo 2018 | Qualitative   | To explore national stakeholders' perceptions of current CPG development activity for primary healthcare (PHC) in South Africa. More specifically,                                                          | South Africa                                                                                                                                                   | Thirty-seven participants from a range of disciplines, stakeholder groups and provinces in the country. The majority were providers of healthcare working | In-depth individual interviews, conducted by two interviewers per interview. The semi-structured interview guide was tailored to the experiences of each interviewee and each interview built on the findings of previous interviews. All interviews were transcribed by a third party, blind to the aims of the study. | Thematic analysis and the lens of an interpretivist conceptual framework. Initial and revised codes were collated into                                                                            |

|                 |                                        |                                                                                                                                                                                                                    |                                                                                                                                                                                 |                                                                                                                                                                                                                                                                                                                                             |                                                                                                                                                                                                                                                                                                                                |                                                                                                                                                                                                                                                                 |
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|                 |                                        | it sought to investigate their perceptions of the processes that should inform national clinical practice guidelines (CPG) development, and their sense of what is actually occurring.                             |                                                                                                                                                                                 | in academia or government.                                                                                                                                                                                                                                                                                                                  |                                                                                                                                                                                                                                                                                                                                | potential themes and produced into an overall 'thematic map' to guide further analyses. Using the thematic map, the themes related to the processes of developing CPGs were identified, named and extracted.                                                    |
| Lamontagne 2021 | Single-blind crossover pragmatic study | To document the acceptability, feasibility, and outcomes of 2 methods (Focus group vs. Online wiki) of involving patients in clinical practice guideline co-design for adults with traumatic brain injuries (TBI). | The study activities took place at the Quebec (Canada) community-based association of individuals with TBI (Association TCC des Deux-Rives) (for the training session and focus | Participants were French-speaking patients with a moderate to severe TBI (Glasgow Coma Scale <13). 16 individuals with TBI participated (8 per group). 11 (68.8%) were male, median age of 44.9 (range 20-57). 10 had a severe traumatic brain injury (62.5%). In terms of living situation, 9 (56.3%) lived alone and 7 (43.4%) lived with | Acceptability was assessed using electronic or paper surveys. The feasibility of the intervention was evaluated using: (1) the number of participants who attended (2) the number of participants who completed the intervention, and (3) the number of support interventions required regarding the focus group and the wiki. | All qualitative data underwent a thematic content analysis based on Sidani and Braden's conceptual framework of acceptability. An interjudge validation process was done by 2 team members to ensure the validity of the analysis until a level of agreement of |

|              |                                     |                                                                                                                   |                                                                    |                                                                                                                                                                                                                                   |                                                                                                                                                                                                                                                           |                                                                                                                                                       |
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|              |                                     |                                                                                                                   | group) and the participant's home (wiki).                          | close relatives. No participants were aware of CPGs prior to the study.                                                                                                                                                           |                                                                                                                                                                                                                                                           | 90% was found in the coding. Divergences in coding were resolved by a third analyst. Themes were validated according to convergences and divergences. |
| Lanza 2000   | Qualitative                         | To develop clinical practice guidelines for a community meeting to prevent and intervene with assaultive behavior | Edith Nourse Rogers Memorial Veterans Hospital, Massachusetts, USA | Thirty-four participants; most were middle-aged White males who were single who had either high-school or college education levels. Most (80%) had been hospitalized for psychiatric reasons 5 times or less in the last 5 years. | Delphi questionnaire and focus group discussion. No further details reported.                                                                                                                                                                             | Patient responses were compared with panelist responses from the second iteration. No further details reported.                                       |
| Lindsay 2020 | Non-randomized Mixed Methods Design | To evaluate the feasibility, acceptability, perceived value, and effectiveness of a new engagement model          | Canada                                                             | Participants collectively had lived experience as a person with a stroke or family member. The first panel was composed                                                                                                           | The authors adapted the Patient and Public Engagement Evaluation Tool (PPEET), a validated public and patient evaluation tool to evaluate the Community Consultation and Review Panel (CCRP) model. Data collection and evaluation included key informant | Not reported.                                                                                                                                         |

|             |                                  |                                                                                                                                                                                                                             |                                                                                                         |                                                                                                                                                                                                                                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                                                                                                                       |
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|             |                                  | in the update of the 2 components of the Canadian Stroke Best Practice Recommendations (CSBPR) Rehabilitation, Recovery and Community Participation module                                                                  |                                                                                                         | of 5 females (mean age: 41.8 years) and 4 males (mean age: 57.3 years). There were 7 patients and 2 caregivers from 7 Canadian provinces. The second panel was composed of 5 females (mean age: 41.8 years) and 3 males (mean age: 52.5 years), representing 6 patients and 2 caregivers from 7 provinces. | interviews, informal discussions and online surveys.                                                                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                                                       |
| Pigott 2021 | Mixed Methods Iterative Approach | To present the Guideline Participant Tool (GPT) to support effective participation in guideline groups, in particular those using the Grading of Recommendations, Assessment, Development, and Evaluation (GRADE) approach. | International (5 continents): Europe n=6, Africa n=1, Americas n=1, Asia n=1, Australia and Pacific n=1 | Interview participants (n=10) Gender: Male n=6, Female n=4; Guideline methodologist n=4, Content Expert n=4, Sponsor Organization n=1, Patient Representative n=1; No Previous Experience: n=2, Experience with 1 guideline n=1, Experience with                                                           | (1) a systematic review of guideline handbooks to identify recommendations for guideline participants.<br><br>(2) interviews with individuals who participated in previous guideline development projects to ascertain their feedback on the tool, GPT.<br><br>(3) used the GPT prospectively in three guideline groups and sought feedback from guideline group chairs, members, and organizers. | (1) Guideline manual review: Three authors identified items for first iteration of the GPT.<br><br>(2) Qualitative interviews: researchers (n=3) independently coded the transcripts using a closed loop iterative process. Conducted |

|             |                         |                                                                                                                                |               |                                                                                                                                                                                                                                                                                                     |                                                                                                                                                                                                                                                                                                                               |                                                                                                                                                                                                 |
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|             |                         |                                                                                                                                |               | <p>more than 1 guideline: n=7</p> <p>Organizations (n=3):</p> <ul style="list-style-type: none"> <li>American Society of Hematologists (ASH)</li> <li>European Commission Initiative on Breast Cancer (ECIBC)</li> <li>WHO's Guideline on tuberculosis infection prevention and control.</li> </ul> |                                                                                                                                                                                                                                                                                                                               | <p>independent thematic analyses of interview transcripts, then met to collate, discuss, and finalize the analysis.</p> <p>(3) External validation survey: Used feedback to refine the tool</p> |
| Pottie 2021 | Iterative mixed methods | To present a structured approach for assessing stakeholder perceptions and implementing the approach in guideline development. | International | Members of guideline development groups from WHO, Canadian Task Force on Preventive Health Care (CTFPHC), and the European Center for Disease Prevention and Control, as well as GRADE methodologists, researchers, and                                                                             | Developed ad hoc in 2015 (pilot). A GRADE Working Group was established. Pretesting with focus groups was conducted. The FACE instrument was used and evaluated with internal and external stakeholders. Refined the instrument using workshops at GRADE Working Group meetings (2017 Rome, Italy, and 2019 Hamilton, Canada) | Not report                                                                                                                                                                                      |

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|                |             |                                                                                                                                                                                                                                 |             | students. External stakeholders included policy makers, physicians, public health workers, health economists, and members of national and international organizations.                                 |                                                                                                                                            |                                                                                                                                                                                                                                                                                                                                             |
| Schalkers 2017 | Qualitative | (1) To develop a tool that could stimulate guideline developers to take children into account on a more structural basis and (2) to explore how to facilitate children's participation in the process of guideline development. | Netherlands | N=47. Professionals involved in guideline development: Patient organizations n=16; Hospitals n=10; Knowledge institutions n=8; Branch associations n=7 ;n= Health funding agencies n=3 and Others n=3. | Three-phased multimethod: (1) Semi-structured interviews with open-ended questions; (2) online questionnaire; (3) focus group discussions. | The interviews were audio recorded and transcribed verbatim for qualitative analysis. A summary was sent to the interviewees to confirm that it properly reflected their views and experiences. All transcripts were read in their entirety and analysed using a combination of deductive and inductive content analysis. The questionnaire |

|             |                               |                                                                                                                                                                                                                                |                                        |                                                                                                                                                                                                                                                                                                                                                                         |                                                                                                                                                                                                                                                                                                                                                                                                                |                                                                    |
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|             |                               |                                                                                                                                                                                                                                |                                        |                                                                                                                                                                                                                                                                                                                                                                         |                                                                                                                                                                                                                                                                                                                                                                                                                | responses were analysed through descriptive statistics using SPSS. |
| Selby 2017  | Qualitative                   | To develop and encourage the adoption of clinical practice guidelines (CPGs) for smoking cessation in Canada by engaging stakeholders in the adaptation of existing high-quality CPGs using principles of the ADAPTE framework | Ontario, Canada                        | <p>There were seven members of the GDG including a family physician, public health researcher, health psychologist and physician specialists from respiratory, addiction and cardiology.</p> <p>The ADAPTE Network included over 800 members. Approx. ⅔ of members were providers of healthcare. End-users such as researchers and policymakers were also included.</p> | Initial review to identify existing clinical practice guidelines, followed by the development and establishment of the ADAPTE network. Workshops, discussions, teleconferences, webinars, and online surveys were conducted to further discuss the guideline, strategies for implementation and to gather feedback on the guideline. The online discussion board allowed continuing feedback on the guidelines | None reported.                                                     |
| Synnot 2022 | Mixed: Scoping review and key | To identify principles and approaches to broaden the                                                                                                                                                                           | International; networks of the project | Two consumers and two guideline developers (three women and one                                                                                                                                                                                                                                                                                                         | An experienced qualitative researcher conducted 60-min interviews over Zoom. Questions probed participants' experiences of engaging consumers/being engaged in                                                                                                                                                                                                                                                 | Conducted a descriptive synthesis of relevant text                 |



|                        |                      |                                                                               |                                                                                                                                               |                                                                                                                                                                                                                                                                                                                                                                                                                                                           |                                                                                                                                                                                                                                       |                                                                                                                                                                                                                                                                                                                                                                |
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|                        | informant interviews | diversity of consumers engaged in guideline development.                      | team and those of international guideline groups, such as the National Institute for Clinical Excellence and Guidelines-International Network | man), based in Europe (n=3) and Australia (n=1). Consumers self-identified as belonging to a minority ethnic group (n=1) and being older (n=2) and had contributed to two or three guidelines. Guideline developers had engaged people with autism and people with a neglected tropical disease. Consumers were experienced as members of a GDG. For one guideline developer, the engagement involved interviews with consumers in low-resource settings. | guidelines, whether they felt able to make an active contribution (consumers only), what worked well, what could have been improved and their recommendations.                                                                        | and transcripts. One researcher undertook line-by-line coding. The authors reviewed the free codes, seeking like concepts, then merged and refined codes before grouping them under sub themes and then themes. The authors used these codes, subthemes and themes as a framework to analyze the interview transcripts. A second researcher reviewed the data. |
| Van de Bovenkam p 2015 | Qualitative          | To study why problems arise in participation practice and to think critically | The Netherlands                                                                                                                               | Professionals and people responsible for the organization of guideline development (n =                                                                                                                                                                                                                                                                                                                                                                   | Analyzed 62 Dutch guidelines developed by the Dutch Council for Quality of Healthcare. The researchers then scored these guidelines using AGREEt. They then conducted semi-structured interviews. Topics discussed included: ideas on | Analyzed by incremental coding. Quotations used have been                                                                                                                                                                                                                                                                                                      |

|                         |             |                                                                                                                                                                                                                                                                        |                 |                                                                                                                                                                                            |                                                                                                                                                                                                  |                                                                                                                                                                                                                                                     |
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|                         |             | about the consequences for future participation practices. This objective addresses the following research question: Are patients involved in the development in Dutch guidelines, what are the experiences with this and how can participation practices be improved? |                 | 15) and representatives of patient organizations (n = 10).                                                                                                                                 | the importance of patient participation, experiences with participation and, based on those, ideas about the future of participation. All the interviews were recorded and transcribed verbatim. | slightly modified only to ensure anonymity and legibility. However, details of analysis methods of the semi-structured interviews were not discussed.                                                                                               |
| Van der Ham 2014 & 2016 | Qualitative | To obtain insights into practices and experiences of service user involvement in mental health guideline development, and extract applicable lessons learned to contribute to approaches for the monitoring and evaluation of such initiatives                         | The Netherlands | N=24. Stakeholders, including: guideline development professionals (n = 4), health care professionals (n = 10), service user representatives (n = 9) and one carer representative (n = 1). | Observations, document analysis, semi-structured interviews, focus group discussions and case studies                                                                                            | Thematic analysis; First, codes were given to features of the data that the researchers considered essential to the research question. A distinction was made between codes explaining the process of service user involvement and codes related to |

|                |                                                  |                                                                                                                                  |                      |                                                                                                                                            |                                                                                                                                                                 |                                                                                                                                                                                                                                                                 |
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|                |                                                  |                                                                                                                                  |                      |                                                                                                                                            |                                                                                                                                                                 | the outcomes of this. Second, core themes were identified and named. These themes explain larger sections of the data by combining different related codes. Third, barriers and facilitators associated with the different themes were identified as sub-themes |
| Van Wersh 2001 | Qualitative                                      | To describe and discuss the experience with four different methods of consumer involvement of evidence based clinical guidelines | North of England, UK | Patient representatives and patients with chronic conditions such as asthma or angina. No further details were reported. (n= not reported) | (1) Guideline development group discussion and individual interviews (2) Forum discussion; (3) Role play, direct observation, workshops (4) telephone interview | No analysis details reported                                                                                                                                                                                                                                    |
| Walker 2022    | Iterative mixed methods participatory case study | To highlight the extensive design process used to develop "Alberta's Home to Hospital                                            | Alberta, Canada      | Over 750 stakeholders, engaged at different stages of the process. This                                                                    | Activities to establish Guideline content: i) learning collaborative; ii) design-team; iii) targeted online surveys; iv) primary care stakeholder               | The learning collaborative was themed using a content analysis approach to                                                                                                                                                                                      |

|  |  |                                                                                                   |  |                                                                                                                                                                                                                                           |                                                                             |                                                                                                                                                                                                                                                                                                                                                                                  |
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|  |  | to Home Transitions Guideline”, with a focus on the participatory design approach used throughout |  | included patients and families, physicians, allied health professionals, provincial leaders, AHS zone operations, Alberta Medical Association (AMA), community and healthcare providers, Strategic Clinical Networks, and Alberta Health. | consultation; v) modified Delphi panel; and vi) patient advisory committee. | identify points of consensus within each guideline element. Focus groups were analyzed to identify key themes across design team member responses. For surveys, finite mixture modeling and ordinal logistic regression were used to determine disagreement within and between groups, respectively. Interviews and focus groups from consultations were transcribed and themed. |
|--|--|---------------------------------------------------------------------------------------------------|--|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|

## **Excluded studies**

We excluded 693 citations after full-text review. Studies were excluded for the following reasons (Figure 4): Did not report on stakeholder engagement (n = 202); not related to guideline development (n=158); wrong study design (e.g., editorials, commentaries) (n=102); reports of single guidelines [e.g., which described the development process but did not empirically explore barriers/facilitators to engagement] (n=102); duplicates (n=16); topics not related to health (n=2), and ‘other’ (e.g., poster abstracts) (n=17). Additionally, 94 citations were excluded because they do not report barriers or facilitators to engaging stakeholders in guideline development, but were considered for inclusion in other reviews in this series (Khabisa 2022; Lytvyn (in press); Petkovic 2022).

## **Methodological quality of included studies**

We critically appraised studies using the CASP checklist for qualitative studies. A summary of the critical appraisal is presented in Figure 5 and Figure 6. All studies, except for one (Carter 1995), reported clear objectives. Generally, the methods selected to collect data were appropriate to collect the subjective experiences of participants (e.g., interviews, focus groups, surveys), but the strategies to recruit these participants were not always explicit or relied on convenience/purposive samples. Therefore, it is possible that the perspectives collected may not be generalizable to all stakeholder groups or all guidelines. In some retrospective case studies (e.g., Chalmers 2017), the data is derived from guideline developer perspectives only (e.g., presented as “lessons learned”). The most common weakness across all studies was a lack of reflexivity/positionality by the study authors. Often, researchers collected data during or after guideline development, and the study authors overlapped with the guideline secretariat (guideline developer) - therefore the authors collecting data may have had prior relationships with the participants, and the authors did not discuss their potential influence on participant responses. In many studies, the reporting of their analytic methods and approval of an ethics review board were unclear.

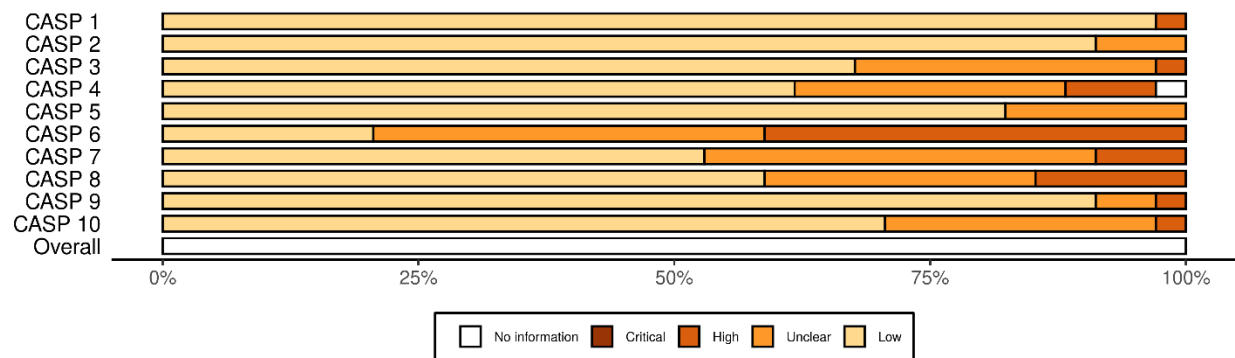


**Figure 5. Methodological quality of all included studies**

CASP Questions:

1. Was there a clear statement of the aims of the research?
2. Is a qualitative methodology appropriate?
3. Was the research design appropriate to address the aims of the research?
4. Was the recruitment strategy appropriate to the aims of the research?
5. Was the data collected in a way that addressed the research issue?
6. Has the relationship between researchers and participants been adequately considered?
7. Have ethical issues been taken into consideration?
8. Was the data analysis sufficiently rigorous?
9. Is there a clear statement of findings?
10. How valuable is the research?

|                       | CASP Critical Appraisal |    |    |    |    |    |    |    |    |     |         |
|-----------------------|-------------------------|----|----|----|----|----|----|----|----|-----|---------|
|                       | D1                      | D2 | D3 | D4 | D5 | D6 | D7 | D8 | D9 | D10 | Overall |
| Armstrong 2017        | +                       | +  | +  | +  | +  | +  | +  | +  | +  | +   |         |
| Armstrong 2019        | +                       | +  | +  | +  | +  | X  | X  | +  | +  | +   |         |
| Armstrong 2020        | +                       | +  | +  | X  | +  | -  | +  | +  | +  | +   |         |
| Atkins 2013           | +                       | +  | -  | -  | +  | X  | +  | +  | +  | -   |         |
| Bartelmann 2020       | +                       | +  | +  |    | +  | -  | -  | +  | +  | +   |         |
| Biggane 2022          | +                       | +  | +  | +  | +  | +  | +  | +  | +  | +   |         |
| Boivin 2010           | +                       | +  | -  | X  | +  | X  | X  | X  | +  | -   |         |
| Brouwers 2017         | +                       | +  | +  | +  | +  | -  | +  | +  | +  | +   |         |
| Brouwers 2018         | +                       | +  | -  | -  | -  | X  | +  | X  | +  | -   |         |
| Carter 1995           | X                       | -  | -  | +  | -  | X  | X  | X  | +  | -   |         |
| Chalmers 2017         | +                       | +  | X  | -  | -  | -  | -  | X  | +  | -   |         |
| Chartres 2020         | +                       | +  | +  | +  | +  | -  | -  | +  | +  | +   |         |
| Daraz/Murad 2019      | +                       | +  | +  | +  | +  | X  | +  | -  | +  | +   |         |
| Den Breejen 2014      | +                       | +  | -  | +  | +  | -  | +  | +  | +  | +   |         |
| Duff 1996             | +                       | +  | -  | -  | +  | X  | -  | X  | +  | +   |         |
| Hameen-Antilla 2016   | +                       | +  | +  | +  | +  | -  | +  | +  | +  | +   |         |
| Harding 2010/2011     | +                       | +  | +  | -  | +  | +  | -  | +  | -  | +   |         |
| Ilott 2006/Rapu 2005  | +                       | +  | -  | +  | -  | X  | +  | +  | +  | -   |         |
| Jarret 2004           | +                       | +  | -  | X  | +  | -  | -  | -  | +  | -   |         |
| Kim 2021              | +                       | +  | +  | +  | +  | +  | +  | +  | +  | +   |         |
| Kopke 2019            | +                       | +  | +  | +  | +  | +  | +  | +  | +  | +   |         |
| Kredo 2018            | +                       | +  | +  | -  | +  | +  | +  | +  | +  | +   |         |
| Lamontagne 2021       | +                       | +  | +  | +  | +  | +  | +  | +  | +  | +   |         |
| Lanza 2000            | +                       | -  | +  | -  | -  | -  | -  | -  | X  | X   |         |
| Lindsay 2020          | +                       | +  | -  | +  | +  | X  | +  | -  | +  | +   |         |
| Pigott 2021           | +                       | +  | +  | +  | +  | X  | +  | +  | +  | +   |         |
| Pottie 2021           | +                       | -  | -  | -  | -  | X  | -  | -  | -  | -   |         |
| Schalkers 2017        | +                       | +  | +  | +  | +  | X  | +  | +  | +  | +   |         |
| Selby 2017            | +                       | +  | +  | +  | +  | -  | -  | -  | +  | +   |         |
| Synot 2022            | +                       | +  | +  | +  | +  | X  | +  | +  | +  | +   |         |
| van de Bovenkamp 2015 | +                       | +  | +  | +  | +  | -  | -  | -  | +  | -   |         |
| van der Ham 2014/2016 | +                       | +  | +  | +  | +  | -  | -  | +  | +  | +   |         |
| van Wersh 2001        | +                       | +  | +  | +  | +  | -  | -  | -  | +  | +   |         |
| Walker 2022           | +                       | +  | +  | -  | +  | X  | -  | -  | +  | +   |         |

D1: CASP 1  
D2: CASP 2  
D3: CASP 3  
D4: CASP 4  
D5: CASP 5  
D6: CASP 6  
D7: CASP 7  
D8: CASP 8  
D9: CASP 9  
D10: CASP 10

**Judgement**  
X High  
- Unclear  
+ Low  
Not applicable

Figure 6. Summary of methodological quality

## **Synthesis of Findings**

We identified a total of a total of 52 findings, encompassing 24 barriers and 28 facilitators to stakeholder engagement in health guideline development. We mapped these findings to the 18 topics of the GIN-McMaster checklist for guideline development and identified barriers for 12/18 topics (Table 5) and facilitators for 14/18 topics (Table 6). Barriers and facilitators were most commonly reported for patients and caregivers, followed by providers of care, principal investigators, and members of the public. Notably, no evidence was available for payers of research.



**Table 5. Barriers to stakeholder engagement in guideline development**

| GIN-McMaster Checklist for Guideline Development      | WHO DOES THE FINDING APPLY TO?                                    | PERSPECTIVE                                                                           | BARRIER                                                                                                                           | EXAMPLE QUOTES                                                                                                                                                                                                                                                                                                                          | STUDIES CONTRIBUTING TO FINDING                                                             | TDF DOMAIN                          |
|-------------------------------------------------------|-------------------------------------------------------------------|---------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------|-------------------------------------|
| <b>1. Organisation, Budget, Planning and Training</b> | Patients, caregivers & patient organisations<br>Providers of care | Patients, caregivers & patient organisations<br>Providers of care                     | 1.1 A lack of human and financial resources at the organisational level limited the collaborative development of recommendations. | <p>“The two project leaders [providers of care] said they underestimated the project coordinator’s workload, particularly in combining the coordination tasks with writing the draft guidelines” (Den Breejen 2014).</p> <p>“There was a lack of infrastructure resources to cope with the logistics of consultations” (Ilott 2006)</p> | Brouwers 2017; Den Breejen 2014; Hameen-Antilla 2016; Ilott 2006/Rapu 2005; Van Wersch 2001 | Environmental context and resources |
|                                                       | Patients<br>Public                                                | Guideline developers<br>Principal investigators<br>Patient and public representatives | 1.2 Guideline development organisations have unclear goals related to patient and public engagement.                              | “Workshop participants [guideline developers, researchers, patients and public] agreed on the importance of patient and public involvement in clinical practice guidelines. However, they reported that patient and public involvement                                                                                                  | Boivin 2010; Lanza 2000                                                                     | Goals                               |

|                                      |          |                                                                                           |                                                                                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                         |                                                  |                                       |
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|                                      |          |                                                                                           |                                                                                    | goals are often largely implicit or articulated in vague terms by their organisations, which makes it difficult to assess success or failure.” (Boivin 2010).                                                                                                                                                                                                                                                                                           |                                                  |                                       |
|                                      | Patients | Guideline developers<br>Representatives of patient organisations<br>Patients & caregivers | 1.3 Guideline developers deliberately exclude patients from guideline development. | “We have deliberately chosen [...] not to involve patients that much. Because then you will get all these chapters on ‘the patient needs to be informed properly’, ‘the patient needs to be supported properly’, ‘the patient needs this’ and ‘the patient needs that’, and that is of course the same for every oncology patient. So we said we are not going to invest time and money in that [within a specific guideline].” (van de Bovenkamp 2015) | Brouwers 2018; Van de Bovenkamp 2015; Selby 2017 | Knowledge                             |
| <b>2. Priority Setting</b>           | -        |                                                                                           | None reported                                                                      |                                                                                                                                                                                                                                                                                                                                                                                                                                                         | -                                                | -                                     |
| <b>3. Guideline Group Membership</b> | Patients | Patients<br>Providers of care<br>Program managers                                         | 3.1 Challenges identifying “representative”                                        | “A characteristic considered a barrier by several professionals and                                                                                                                                                                                                                                                                                                                                                                                     | Armstrong 2020; Biggane 2022; Brouwers 2018;     | Social/Professional Role and Identity |

|  |          |                                                                    |                                                                                                                               |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                       |                                                                                                           |                                       |
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|  |          | Guideline developers<br>Author observations                        | patients to engage in guideline development.                                                                                  | service user representatives is the tendency of certain service users to focus merely on their personal experiences with illness and care.” (van der Ham 2014)                                                                                                                                                                                                                                                                                                                                                                        | Chalmers 2017; Hameen-Antilla 2016; Kim 2021; Lanza 2000; Van Der Ham 2014; van der Ham 2016; Walker 2022 |                                       |
|  | Patients | Patients & caregivers<br>Providers of care<br>Guideline developers | 3.2 Patients perceive ‘tokenistic engagement’ which limits their ability to meaningfully contribute to guideline development. | “Some concern was expressed that the inclusion of service users was tokenistic and that this was evidenced by there being an average of only two service users compared to six to eight healthcare professional members serving on advisory groups: ‘Ah, now then. I think the, um, service-user representation is a bit tokenistic’. (Healthcare professional); ‘I think it says something about how much experts by experience are valued, because if we’re all equally valued, there’d be equal numbers of people’.” (Atkins 2013) | Armstrong 2017; Atkins 2013; Biggane 2022; Brouwers 2017; Daraz 2019; Jarret 2004; Murad 2019;            | Social/Professional Role and Identity |

|                                                          |                                                                |                                                                                    |                                                                                                                                         |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |                                                                                                                  |                                          |
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|                                                          | Patients<br>Public                                             | Patients<br>Public<br>Guideline<br>developers                                      | 3.3 Language fluency<br>may limit the<br>representativeness of<br>patients and their<br>ability to engage in<br>group processes.        | “Interviewees also noted<br>from their experience that<br>differences in cultures<br>and languages can have a<br>major impact on group<br>process and that respect<br>and consideration for this<br>should be given by all<br>guideline participants.”<br>(Piggott 2021)                                                                                                                                                                                                                                         | Armstrong 2017;<br>Chalmers 2017; Piggott<br>2021                                                                | Skills                                   |
| <b>4. Establishing<br/>Guideline Group<br/>Processes</b> | Patients<br>Providers of<br>care<br>Principal<br>investigators | Patients<br>Providers of care<br>Program<br>Managers<br>Principal<br>Investigators | 4.1 Unclear roles and<br>responsibilities for<br>guideline<br>development group<br>members may<br>weaken or disrupt<br>group processes. | “The evidence review<br>groups members felt that<br>many of the tensions that<br>were experienced<br>between groups of<br>participants could have<br>been limited if working<br>committees were<br>provided with clear<br>information about the<br>different roles of the 2<br>groups.” (Chartres 2020)<br><br>“Some patient<br>representatives were<br>uncertain about what was<br>expected of them because<br>the roles of patient<br>representatives were not<br>explicitly discussed.”<br>(van der Ham 2016) | Carter 1995; Chartres<br>2020; Jarret 2004;<br>Lanza 2000; Kim<br>2021; Van Der Ham<br>2014; Van Der Ham<br>2016 | Social/Professional<br>Role and Identity |

|  |                               |                                                             |                                                                                                                                       |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |                                                                    |                            |
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|  | Patients<br>Providers of care | Patients<br>Providers of care<br>Principal<br>investigators | 4.2 Failing to manage power imbalances between patients and providers of care may affect group dynamics and limit patient engagement. | <p>“Most members thought that discussion was dominated by some to the exclusion of other members, in particular service users: ‘I don’t remember many items we’ve discussed where I’ve heard a patient’s perspective sought or, or expressed’. (Healthcare professional) ‘It did strike me ... that there are ... some people who are more voluble and find it easier to get space on the floor than others’.</p> <p>(Healthcare professional)...‘I think there is a hierarchy of opinion-formers within the group... I think the hospital physicians would come nearer the top of it... Probably the ... patient representatives [at the bottom]’. (Healthcare professional)” (Atkins 2013)</p> | Armstrong 2017; Atkins 2013; Carter 1995; Harding 2010; Kredo 2018 | Social influences          |
|  | Patients                      | Patients<br>Principal<br>Investigators<br>Providers of care | 4.3 A lack of confidence in their skills and legitimacy may reduce patient                                                            | “Furthermore, it was evident that some patients were more confident and                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                          | Armstrong 2017; Brouwers 2018; Chalmers 2017;                      | Beliefs about capabilities |

|                                                           |          |                                                                                             |                                                                                                                      |                                                                                                                                                                                                                                                                                                                                                         |                                                                                                           |                   |
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|                                                           |          | Program managers                                                                            | members' level of engagement.                                                                                        | comfortable than others in contributing during meetings.” (Brouwers 2018)<br><br>“Participants compared themselves with other GDG members, which could undermine confidence: ‘I thought ‘here are all these very professional people, the experts . . . and there’s little old me that’s had this experience and it was a long time ago” (Harding 2010) | Harding 2010; Kim 2021                                                                                    |                   |
| <b>5. Identifying Target Audience and Topic Selection</b> | -        |                                                                                             | None reported                                                                                                        |                                                                                                                                                                                                                                                                                                                                                         | -                                                                                                         | -                 |
| <b>6. Consumer and Stakeholder Involvement</b>            | Patients | Patients and caregivers<br>Providers of care<br>Principal investigators<br>Program managers | 6.1 Patients’ engagement may be limited by feelings of intimidation derived from the process or other group members. | “Participants [cancer patients, survivors, family members and caregivers] agreed that feeling intimidated may be a barrier that prevents patients from participating on practice guideline committees. Intimidation occurs because many non-patient members of these                                                                                    | Armstrong 2017; Atkins 2013; Brouwers 2017; Brouwers 2018; Duff 1996; Harding 2010; Jarret 2004; Kim 2021 | Social influences |

|                                               |          |          |                                                                                                                               |                                                                                                                                                                                                                                                                                                                                                                                                                                                                              |                                              |                            |
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|                                               |          |          |                                                                                                                               | committees are highly trained healthcare professionals. Therefore, patients may feel uncomfortable commenting or questioning a point being made by one of the committee members.” (Brouwers 2017)                                                                                                                                                                                                                                                                            |                                              |                            |
| <b>7. Conflict of Interest Considerations</b> | Patients | Patients | 7.1 Patient engagement may be affected by concerns around confidentiality and judgement in the guideline development process. | <p>“Several individuals expressed discomfort at the idea of participating with their personal physician, fearing loss of confidentiality or judgement regarding their opinions” (Armstrong 2017)</p> <p>“Patient confidentiality should be maintained at all times. Patients may choose to share information about their medical background, but their physician should not disclose information to the panel or panel members without patient consent.” (Chalmers 2017)</p> | Armstrong 2017; Brouwers 2017; Chalmers 2017 | Beliefs about consequences |

|                                      |                                 |                                            |                                                                                                                                     |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                         |                          |                                       |
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| <b>8. (PICO) Question Generation</b> | Patients<br>Public              | Patients<br>Public                         | 8.1 Patients and the public perceived that health outcome selection is outside of their purview.                                    | “[...] lay members largely did not recall the focus on the importance of their role in health outcome selection. When asked, [...] most did not mention health outcomes as an area they were, or even wanted to be involved with [...]. The only lay member who recalled learning about evidence review protocols and health outcome selection [...] characterised the protocol setting and thus, health outcome selection as something that was outside her remit, as a: ‘system the technical team would go through’.” (Biggane 2022) | Biggane 2022             | Social/Professional Role and Identity |
|                                      | Patients & caregivers<br>Public | Guideline developers<br>Author observation | 8.2 Time constraints and inflexible timelines may limit patient and public engagement in outcome selection and question generation. | “The data indicated that lay members and health professionals needed time to familiarise themselves with the guideline development process and with each other. As we show below, the lack of such time restricted the input                                                                                                                                                                                                                                                                                                            | Biggane 2022; Kopke 2019 | Environmental context and resources   |



|                                                                                                   |                               |                                                                                  |                                                                                                                                               |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                      |                                                                                      |                    |
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|                                                                                                   |                               |                                                                                  |                                                                                                                                               | of lay members in health outcome selection.” (Biggane 2022)                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                          |                                                                                      |                    |
| <b>9. Considering Importance of Outcomes and Interventions, Values, Preferences and Utilities</b> | Patients<br>Providers of care | Providers of care<br>Principal Investigators<br>Patients<br>Guideline developers | 9.1 Power imbalances between patients and providers of care in guideline development groups may promote unequal influence in decision-making. | <p>“There was also a perceived inequality between members’ contributions when it came to influencing decision making: ‘I think you would have to shout quite loud to say that you had equal influence’.</p> <p>(Service user) [...] There were twice as many comments about there being unequal contributions of members to discussion and influence on subsequent decision making than comments describing equal contributions of members. While this was the case among service users and health professionals, advisory group chairs were more likely to say there were equal contributions to discussion and decision making.” (Atkins 2013)</p> | Atkins 2013; Harding 2010; Jarret 2004; Lanza 2000; Rapu 2005; Van de Bovenkamp 2015 | Decision Processes |

|                                                                         |                   |                                                                                       |                                                                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                                                      |                                                                                                                             |           |
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| <b>10. Deciding what Evidence to Include and Searching for Evidence</b> | Patients          | Patients & patient organisations<br>Providers of care<br>Guideline developers         | 10.1 Patient engagement in guideline development may be limited by a lack of technical skills applicable to reviewing scientific evidence. | “While some patients may be able to participate in the evidence review and in the creation of a final guideline document, this is dependent on the topic of the guideline and the patients’ individual levels of expertise and skill.” (Brouwers 2017)                                                                                                                                                                               | Brouwers 2017; Hameen-Antilla 2016; Kredo 2018; Schalkers 2017; Van De Bovenkamp 2015; Van Der Ham 2014                     | Skills    |
|                                                                         | Patients          | Patients<br>Providers<br>Payers/Purchasers of health services<br>Guideline developers | 10.2 The impact and depth of patient engagement may be limited by a devaluation of experiential knowledge.                                 | “Interviewees acknowledged that there is little attention paid to the inclusion of children’s perspectives in current guidelines[...] several participants believed that it is due to the hierarchical health care system in which the medical knowledge of professionals is considered as superior to experiential knowledge of patients and that this might be even more the case with children than with adults (Schalkers 2017). | Armstrong 2020; Atkins 2013; Harding 2011; Schalkers 2017; Selby 2017; Van de Bovenkamp 2015; Van der Ham 2014; Walker 2022 | Knowledge |
|                                                                         | Providers of care | Providers of care<br>Principal investigators                                          | 10.3 Diverging disciplines and approaches between                                                                                          | “These divergent roles and epistemological beliefs led to conflict and                                                                                                                                                                                                                                                                                                                                                               | Chartres 2020                                                                                                               | Knowledge |

|                                                                        |                                              |                                              |                                                                                                                                                                   |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                              |               |           |
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|                                                                        | Principal investigators                      |                                              | ‘evidence review groups’ (technical teams) and guideline development groups may promote intergroup conflict and division.                                         | division between the groups. Many of the working committee members believed that in order to best protect the public’s health, the evidence presented by the review groups should at times be challenged. [...] The evidence reviews groups however, saw this process as being hostile, aggressive and at times they felt victimised for doing their job. They felt that the evidence-review role they had been contracted to complete was not respected by some working committee members.” (Chartres 2020) |               |           |
| <b>11. Summarizing Evidence and Considering Additional Information</b> | Providers of care<br>Principal investigators | Providers of care<br>Principal investigators | 11.1 Stakeholder engagement may be restricted by a separation of duties and limited communication between guideline development groups and evidence review groups | “The evidence review group [...] members were unable to share with the working committee insights and opinions on the best approach to identifying or evaluating the evidence. Further, the 2 groups were unable to help one another to understand each other’s                                                                                                                                                                                                                                              | Chartres 2020 | Knowledge |

|                                                                         |                   |                                              |                                                                                                              |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                |               |           |
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|                                                                         |                   |                                              |                                                                                                              | point of view. The transfer of ideas, knowledge and expertise was limited by the separation of the 2 groups [...] This separation between the groups was also seen by the evidence review groups as a major limitation in how the guideline process was conducted [...] The evidence review groups felt that this criticism from the working committees grew from the separation of duties, and failure to have effective communication strategies in place. ” (Chartres 2020) |               |           |
| <b>12. Judging Quality, Strength or Certainty of a Body of Evidence</b> | Providers of care | Providers of care<br>Principal investigators | 12.1 Stakeholder engagement in evaluating evidence may be limited by an inadequate understanding of methods. | “Both the working committee and evidence review groups viewed understanding the methods as a significant challenge to the current process. Inadequate understanding of the methods restricts the level of input certain working committee                                                                                                                                                                                                                                      | Chartres 2020 | Knowledge |

|                                                                      |                    |                                                                    |                                                                                                                                |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                             |                                                                                                                                                                                                                            |                    |
|----------------------------------------------------------------------|--------------------|--------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------|
|                                                                      |                    |                                                                    |                                                                                                                                | members can have in the process and creates tensions between the groups.” (Chartres 2020)                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                                                                                                            |                    |
| <b>13. Developing Recommendations and Determining their Strength</b> | Patients<br>Public | Providers of care<br>Patients<br>Public<br>Guideline<br>developers | 13.1 Patient and public engagement may be limited by their lack of content knowledge, clinical expertise and technical skills. | <p>“At the same time, some participants felt that having some content knowledge would be beneficial: ‘As consumers. . . we have to have some knowledge of what is stage 1, what is stage 2, what is stage 3. . . .’” (Armstrong 2017)</p> <p>“Participants felt that [...] it may be too late to consider patient input during the creation of a final document, as the most important decisions and recommendations are finalized by this point. While some patients may be able to participate in the evidence review and in the creation of a final guideline document, this is dependent on the topic of the guideline and the patients’ individual levels of expertise and skill.” (Brouwers 2017)</p> | <p>Armstrong 2017;<br/>Armstrong 2020;<br/>Biggane 2022;<br/>Brouwers 2017;<br/>Brouwers 2018;<br/>Chalmers 2017, Den Breejen 2014; Harding 2011; Selby 2017; Shalkers 2017; Van Der Bovenkamp 2015; Van Der Ham 2014;</p> | Skills & Knowledge |

|  |                                            |                                                      |                                                                                                                             |                                                                                                                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                                              |                                     |
|--|--------------------------------------------|------------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------------|
|  | Patients & caregivers<br>Providers of care | Patients & caregivers<br>Providers of care           | 13.2 Insufficient and competing demands of time may limit stakeholder engagement in recommendation development.             | “Some members thought there was not enough time to complete recommendations, with a sense of being rushed towards the end: ‘The actual wording of the recommendations struck me as a little bit rushed and a bit of a free for all towards the end’. (Healthcare professional) ‘There was a bit of steamrolling going on. We were, more or less, accepting things, because there was no time left to discuss it properly’. (Chair)” (Atkins 2013) | Armstrong 2017; Atkins 2013; Bartelmann 2020; Brouwers 2017; Brouwers 2018; Chalmers 2017; Den Breejen 2014; Duff 1996; Ilott 2006; Kredo 2018; Piggott 2021 | Environmental context and resources |
|  | Patients & caregivers<br>Providers of care | Patients<br>Provides of care<br>Guideline developers | 13.3 Unequal access to information technology resources may limit stakeholders’ ability to engage in guideline development. | “One PCM [patient/carer member] had a real problem carrying out her work because she had no printer and hard copies were not supplied.” (Jarrett 2004)                                                                                                                                                                                                                                                                                            | Jarret 2004; Kredo 2018; Synnot 2022                                                                                                                         | Environmental context and resources |
|  | Patients<br>Public                         | Patients<br>Public                                   | 13.4 Technical concepts and professional jargon may negatively impact patient and                                           | “A further issue in lay members’ involvement related to the highly technical nature of the language used by the                                                                                                                                                                                                                                                                                                                                   | Armstrong 2020; Biggane 2022; Brouwers 2018; Harding 2010; Jarret                                                                                            | Knowledge                           |

|                                                                                                       |                                                                          |                      |                                                                                                                                   |                                                                                                                                                                                    |                              |                                     |
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|                                                                                                       |                                                                          |                      | public comfortability and ability to contribute to the guideline development process.                                             | committees and in the literature provided.[...] Lay members unsurprisingly felt the technical nature of discussions inhibited them from participating.” (Biggane 2022)             | 2004; Van Wersch 2001        |                                     |
| <b>14. Wording of Recommendations and of Considerations of Implementation, Feasibility and Equity</b> | -                                                                        |                      | None reported                                                                                                                     |                                                                                                                                                                                    | -                            | -                                   |
| <b>15. Reporting and Peer Review</b>                                                                  | -                                                                        |                      | None reported                                                                                                                     |                                                                                                                                                                                    | -                            | -                                   |
| <b>16. Dissemination and Implementation</b>                                                           | -                                                                        |                      | None reported                                                                                                                     |                                                                                                                                                                                    | -                            | -                                   |
| <b>17. Evaluation and Use</b>                                                                         | -                                                                        |                      | None reported                                                                                                                     |                                                                                                                                                                                    | -                            | -                                   |
| <b>18. Updating</b>                                                                                   | Patients<br>Providers of care<br>Policymakers<br>Principal investigators | Guideline developers | 18.1 Guideline developers may face challenges engaging stakeholders in guideline updates due to insufficient financial resources. | “...when guidelines are updated, time and budget available for the development process is generally limited, creating the risk that less attention will be devoted to service user | Selby 2017; van der Ham 2014 | Environmental context and resources |

|  |  |  |  |                                  |  |  |
|--|--|--|--|----------------------------------|--|--|
|  |  |  |  | involvement.” (van der Ham 2014) |  |  |
|--|--|--|--|----------------------------------|--|--|

**Table 6. Facilitators to stakeholder engagement in guideline development**

| <b>GIN-McMaster Checklist for Guideline Development</b> | <b>WHO DOES THE FINDING APPLY TO?</b>      | <b>PERSPECTIVE</b>                                                 | <b>FACILITATOR</b>                                                                                                                  | <b>EXAMPLE QUOTE</b>                                                                                                                                                                                                                                                                                                                                                                                                                                                                  | <b>STUDIES</b>                                                                                       | <b>TDF DOMAIN</b>                   |
|---------------------------------------------------------|--------------------------------------------|--------------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------|-------------------------------------|
| <b>1. Organisation, Budget, Planning and Training</b>   | Patients & caregivers<br>Providers of care | Guideline developers<br>Patients & caregivers<br>Providers of care | 1.1 Guideline development<br>organisations should<br>Implement formal stakeholder engagement models into their everyday operations: | “The model provided members with opportunities to regularly review and offer feedback on draft recommendations from the scientific writing group, provide context related to their values and preferences in stroke care to help inform what person-centered care meant in the context of their lived experiences. This is a significant advancement from previous models where engagement of people with lived experience as members of the writing groups was minimal and they were | Grant 2021; Ilott 2006/Rapu 2005; Lindsay 2020; Van der Ham 2015; van de Bovenkamp 2015; Walker 2022 | Environmental context and resources |



|  |                                         |                                                                                    |                                                                                          |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                |                                                                                                                                                                                                   |                                                     |
|--|-----------------------------------------|------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------------------------------------------|
|  |                                         |                                                                                    |                                                                                          | often disengaged” (Lindsay 2020).                                                                                                                                                                                                                                                                                                                                                                                                                                              |                                                                                                                                                                                                   |                                                     |
|  | Patients<br>Public<br>Providers of care | Guideline developers<br>Patients & caregivers<br>Public<br>Principal investigators | 1.2 Provide stakeholders with financial compensation and/or professional accreditations: | “The cost of [face-to-face meetings] is the requirement for patients to travel to meetings, which is expensive and time consuming. It is essential that the project funds patient travel adequately and in an appropriate way and timeline” (Chalmers 2017).                                                                                                                                                                                                                   | Armstrong 2017; Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Jarret 2004; Rapu 2005; Synnot 2022                                                                          | Environmental context and resources & Reinforcement |
|  | Patients<br>Public                      | Patients<br>Public<br>Guideline developers<br>Providers of care                    | 1.3 Guideline developers should provide training for patient and public stakeholders.    | “...individuals felt that an orientation session to introduce them to PGs and to describe their role as patient representatives would have aided their understanding of the process. Several participants suggested that minimal formal training, if any, would be required to prepare patients for their involvement in PG development and implementation because, as patient representatives, their role is to represent the opinions, needs and concerns of other patients, | Armstrong 2020; Biggane 2022; Boivin 2010; Brouwers 2017; Chalmers 2017; Chartres 2020; Daraz 2019; Murad 2019; Hameen-Antilla 2016; Duff 1996; Jarret 2004; Brouwers 2018; Van de Bovenkamp 2015 | Knowledge & Skills                                  |

|                                      |                    |                                                                       |                                                                                                                        |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                          |                                                                                                                                                                                       |                                        |
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|                                      |                    |                                                                       |                                                                                                                        | and not to provide medical or technical expertise.” (Brouwers 2017)                                                                                                                                                                                                                                                                                                                                                                                                                      |                                                                                                                                                                                       |                                        |
| <b>2. Priority Setting</b>           | -                  |                                                                       | None reported                                                                                                          |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                          | -                                                                                                                                                                                     | -                                      |
| <b>3. Guideline Group Membership</b> | Patients<br>Public | Patients<br>Public<br>Principal Investigators<br>Guideline developers | 3.1 Patient/public and wider stakeholder selection should consider the representativeness and diversity of membership. | <p>“Participants highlighted the importance of recruiting patient/public members early in the process, with consideration given to a balanced socio-demographic representation, because many CPG disproportionately impact certain subgroups.” (Boivin 2010)</p> <p>“This study included several guideline topics and participants from diverse backgrounds thus allowing us to analyse the experience of different groups of participants involved in the process.” (Chartres 2020)</p> | Armstrong 2017; Armstrong 2020; Boivin 2010; Brouwers 2018; Chalmers 2017; Chartres 2020; Den Breejen 2014; Grant 2021; Harding 2010; Van Der Ham 2014; Van Wersch 2001; Synnott 2022 | Social/Professional Role and Identity  |
|                                      | Patients           | Patients<br>Providers of care<br>Program managers                     | 3.2 The recruitment of patient representatives should prioritise the identification of knowledgeable and               | “In addition, previous experience of guideline development or other situations in which service user representatives                                                                                                                                                                                                                                                                                                                                                                     | Armstrong 2017; Biggane 2022; Brouwers 2018; Hameen-Anttila 2016; Kim 2021;                                                                                                           | Beliefs about Capabilities & Knowledge |

|  |                               |                                                                                                      |                                                                                                                                       |                                                                                                                                                                                                                                                                                                                                                                     |                                          |                    |
|--|-------------------------------|------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------|--------------------|
|  |                               | Guideline developers                                                                                 | experienced patient representatives.                                                                                                  | interact with professionals can contribute to developing the professional skills and qualities needed in guideline development.” (Van der Ham 2014)                                                                                                                                                                                                                 | Van der Ham 2014;                        |                    |
|  | Patients<br>Providers of care | Patients                                                                                             | 3.3 Guideline developers should consider the ratio and diversity characteristics of patients and physicians within a guideline panel. | “One area of considerable differences of opinion was regarding the optimal number of patients and physicians on the panel, particularly the ratio of the two. [...]: ‘If I was in a group like that I would want to have peers to feel comfortable and I would also want to have probably a balance between people like physicians, authorities.’” (Armstrong 2017) | Armstrong 2017                           | Social influences  |
|  | Principal investigators       | Principal Investigators<br>Providers of care<br>Program managers<br>Patients<br>Guideline developers | 3.4 Guideline development benefits from the inclusion of methodological experts.                                                      | “A few participants identified the need to involve individuals with expertise in research methods to support systematic reviews of research describing patient preferences and the conduct of qualitative interviews or focus groups                                                                                                                                | Chartres 2020;<br>Kim 2021; Piggott 2021 | Knowledge & Skills |

|                                                  |                                                                                                                                     |                                                                                  |                                                                                                                                                                                |                                                                                                                                                                                                                                                                                                                                                                                   |                                                                                                                                          |                                       |
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|                                                  |                                                                                                                                     |                                                                                  |                                                                                                                                                                                | to explore patient preferences.” (Kim 2021)                                                                                                                                                                                                                                                                                                                                       |                                                                                                                                          |                                       |
|                                                  | Patients<br>Providers of care<br>Payers of health services<br>Payers of health research<br>Policymakers<br>Principal investigators. | Providers of care<br>Patients                                                    | 3.5 Taking a network approach to guideline development benefits the engagement of stakeholders in the guideline development process.                                           | “This study provides detailed insight into the feasibility of a novel patient-centered network approach to MuG [multidisciplinary guideline] development for fertility care. This approach enabled the collaborative development of a harmonized set of one MuG and four MoGs [monodisciplinary guidelines] for clinical care pathways for infertile couples.” (Den Breejen 2014) | Carter 1995; Den Breejen 2014; Jarret 2004; Ilott 2006; Rapu 2005; Van der Ham 2014                                                      | Environmental context and resources   |
| <b>4. Establishing Guideline Group Processes</b> | Patients<br>Principal investigators                                                                                                 | Patients<br>Principal investigators<br>Providers of care<br>Guideline developers | 4.1 Clear roles and expectations for stakeholder involvement in guideline development facilitate meaningful engagement, particularly for patients and principal investigators. | “The professional participants in a task force or research project should understand what the role of patients in the project is, and patients should understand exactly what is expected of them.” (Chalmers 2017)                                                                                                                                                               | Boivin 2010; Brouwers 2017; Brouwers 2018; Carter 1995; Chalmers 2017; Chartres 2020; Den Breejen 2014; Duff 1996; Lanza 2000; Rapu 2005 | Social/Professional Role and Identity |
|                                                  | Patients                                                                                                                            | Patients<br>Providers of care                                                    | 4.2 Guideline developers should foster group                                                                                                                                   | “NICE Public Involvement Programme (PIP) facilitate                                                                                                                                                                                                                                                                                                                               | Armstrong 2017; Armstrong 2019;                                                                                                          | Behavioural regulation                |

|  |     |                                         |                                                                                                                                      |                                                                                                                                                                                                                                                                                                                                                                                                                                                                            |                                                                                                                         |                                     |
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|  |     |                                         | processes that promote patient empowerment.                                                                                          | <p>training sessions, to introduce lay members to the guideline development process,[...] When interviewed, lay members largely described this training as “helpful” and “empowering”, praising the way their role, the process and the scientific terminology were explained.” (Biggane 2022)</p> <p>“To empower patient representatives and facilitate meaningful engagement, we developed a curriculum based on content from the GRADE Working Group.” (Murad 2019)</p> | Biggane 2022; Brouwers 2017; Chalmers 2017; Ilott 2006; Jarret 2004; Murad 2019; Van Wersch 2001                        |                                     |
|  | All | Patients<br>Providers of care<br>Public | 4.3 Allowing stakeholders to identify their preferred mode of participation can facilitate more equitable and accessible engagement. | <p>“Online methods facilitate engagement for people with physical or social disabilities, those in geographically dispersed areas in mostly high-income countries, and young people” (Synnot 2022).</p>                                                                                                                                                                                                                                                                    | Armstrong 2017; Armstrong 2019; Bartelmann 2020; Brouwers 2018; Chalmers 2017; Lamontagne 2021; Selby 2017; Synnot 2022 | Environmental Context and Resources |

|  |                                                                                          |                                                                                                                             |                                                                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                        |                                                                                                                                 |                                                           |
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|  | <p>Patients &amp; caregivers</p> <p>Providers of care</p> <p>Principal investigators</p> | <p>Patients</p> <p>Providers of care</p> <p>Principal investigators</p> <p>Program managers</p> <p>Guideline developers</p> | <p>4.4 Involving a skilled moderator/chair in the guideline development process can improve group processes and facilitate engagement.</p> | <p>“Facilitators should be sensitive, skilled moderators, and able to engage all panel members, possibly with pre-established plans for soliciting views, particularly if panel members are hesitant to volunteer opinions.” (Armstrong 2017)</p> <p>“Our findings suggest the chair of a committee can serve as a bridge between lay members and health professionals by inviting and facilitating input from the lay perspective. Thus, it is essential that committee chairs have the appropriate skills, support and training.” (Biggane 2022)</p> | <p>Armstrong 2017; Biggane 2022; Brouwers 2018; Chartres 2020; Duff 1996; Harding 2010; Jarret 2004; Kim 2021; Piggott 2021</p> | <p>Social/Professional Role and Identity &amp; Skills</p> |
|  | <p>All</p>                                                                               | <p>Patients</p> <p>Providers of care</p> <p>Principal investigators</p> <p>Program managers</p>                             | <p>4.5 The provision of practical, technical and emotional support is a facilitator of engagement for all stakeholders.</p>                | <p>“Consumers need to be prepared and supported through training and ongoing support, with many potential meeting adaptations such as using understandable language in meetings and paperwork.” (Synnot 2022)</p>                                                                                                                                                                                                                                                                                                                                      | <p>Chartres 2020; Den Breejen 2014; Duff 1996; Ilott 2006; Jarret 2004; Kim 2021; Selby 2017; Synnot 2022; Van der Ham 2014</p> | <p>Environmental context and resources</p>                |

|                                                           |          |                      |                                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                   |            |       |
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| <b>5. Identifying Target Audience and Topic Selection</b> | Patients | Guideline developers | 5.1 Online Delphi methods facilitate patient engagement in topic selection and priority setting exercises. | <p>“At the beginning of the CPG development process, patients can help nominate and prioritize topics that they find important. Online modified-Delphi processes can be used to have patients systematically propose and develop consensus on the most important CPG topics. This process could either help identify an entire disease state that requires CPGs or decide which topics within that disease state to address. Specifically, patients could propose potential topics in an initial and open-ended “brainstorming” round, and then prioritize them during subsequent rounds using close-ended rating or ranking questions. Alternatively, an online modified-Delphi process could help engage patients in prioritizing an existing list of potential topics.”</p> <p>(Grant 2021</p> | Grant 2021 | Other |
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| <b>6. Consumer and Stakeholder Involvement</b> | Patients                      | Providers of care<br>Principal Investigators<br>Patients<br>Policymakers<br>Guideline developers | 6.1 Patients should be meaningfully involved throughout the guideline development process.               | “Patient involvement in the design, conduct, implementation and dissemination of clinical research and clinical guidelines can greatly enhance the quality and impact of such projects.” (Chalmers 2017)                                                                                                                                                                                                                                                                                                                                                                             | Atkins 2013; Biggane 2022; Brouwers 2017; Brouwers 2018; Chalmers 2017; Jarret 2004; Selby 2017; Van Der Ham 2014 and 2016; | Social/Professional Role and Identity |
| <b>7. Conflict of Interest Considerations</b>  | Patients<br>Providers of care | Patients<br>Guideline developers<br>Providers of care<br>Principal investigators<br>Policymakers | 7.1 Guideline developers should manage conflicts of interest and patient concerns around confidentiality | “Patient confidentiality should be maintained at all times. Patients may choose to share information about their medical background, but their physician should not disclose information to the panel or panel members without patient consent.” (Chalmers 2017)<br><br>“All network members and GDG Section Leads had to disclose any potential or perceived conflicts of interest, especially with pharmaceutical companies. Members of the GDG without commercial conflicts were asked to be Section Leads and adjudicate the final recommendations. In addition, we ensured that | Armstrong 2017; Chalmers 2017; Grant 2021; Selby 2017                                                                       | Beliefs about consequences            |



|                                      |                    |                                                                    |                                                                                                                    |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |              |                                     |
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|                                      |                    |                                                                    |                                                                                                                    | having a majority without conflicts in each section subgroup diluted the effects of any potential commercial bias.” (Selby 2017)                                                                                                                                                                                                                                                                                                                                                 |              |                                     |
| <b>8. (PICO) Question Generation</b> | Public<br>Patients | Public<br>Patients<br>Principal investigators<br>Providers of care | 8.1 Patient and public engagement may benefit from the provision of training specific to health outcome selection. | “Training of all committee members in the importance of health outcome selection could also further strengthen lay member involvement in this area.[...] we have identified several processes which help to facilitate and support their involvement,[...] This includes the importance of appropriate and relevant language, engagement and invitation to participate, training and support for all committee members and considering the development timeline.” (Biggane 2022) | Biggane 2022 | Knowledge & Skills                  |
|                                      | Patients<br>Public | Patients<br>Public                                                 | 8.2 Guideline developers should consider establishing flexible processes and timelines.                            | “Guideline developers should consider more flexible timelines and methods to support committee members.” (Biggane 2022)                                                                                                                                                                                                                                                                                                                                                          | Biggane 2022 | Environmental context and resources |

|                                                                                                   |                                                          |                                            |                                                                                                                            |                                                                                                                                                                                                                                                                                                                                                                                                           |                             |                                       |
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|                                                                                                   | Patients                                                 | Guideline developers<br>Author observation | 8.3 Guideline developers should use empirical methods for engaging with patients.                                          | “Online modified-Delphi processes can be used to have patients systematically propose and develop consensus on the most important CPG topics.” (Grant 2021)                                                                                                                                                                                                                                               | Biggane 2022;<br>Grant 2021 | Other                                 |
|                                                                                                   | Patients<br>Providers of care<br>Principal investigators | Guideline developers                       | 8.4 Guideline development should involve ‘Champions’ and representatives from disease-specific societies or organisations. | “A crucial role in this process was played by the National Multiple Sclerosis Society of each participating country, and particularly by a task force member who was an MS patient and also a member of the MS International Federation, and of the Danish national MS society. She was key in bridging the issues of the MS patients to those of the health professionals and researchers.” (Kopke 2019) | Kopke 2019                  | Social/Professional Role and Identity |
| <b>9. Considering Importance of Outcomes and Interventions, Values, Preferences and Utilities</b> | -                                                        |                                            | None reported                                                                                                              |                                                                                                                                                                                                                                                                                                                                                                                                           | -                           | -                                     |

|                                                                         |                                              |                                              |                                                                                                                                                |                                                                                                                                                                                                                                                                                                                                    |                                                     |                    |
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| <b>10. Deciding what Evidence to Include and Searching for Evidence</b> | Patients                                     | Patients<br>Providers of care                | 10.1 Guideline developers should elicit patient knowledge (experiential evidence) beyond guideline development group membership.               | ““It would have been unthinkable not to have them. In my work we involve consumers all the time in all our projects from conception to completion...The consumers at all times bring you back to the experiences of individual women. Consumers are not an add-on part. They’re an essential part of the process.”” (Jarrett 2004) | Armstrong 2017;<br>Hameen-Antilla 2016; Jarret 2004 | Knowledge          |
| <b>11. Summarizing Evidence and Considering Additional Information</b>  | -                                            |                                              | None reported                                                                                                                                  |                                                                                                                                                                                                                                                                                                                                    | -                                                   | -                  |
| <b>12. Judging Quality, Strength or Certainty of a Body of Evidence</b> | Providers of care<br>Principal investigators | Providers of care<br>Principal investigators | 12.1 Providing training on the frameworks employed in assessing the body of evidence may facilitate the engagement of guideline group members. | “[...] more pragmatic advice and training by the NHMRC for the working committee members unfamiliar with the frameworks used to assess the body of evidence is necessary, not only at the commencement of the process but should be ongoing thereafter.” (Chartres 2020)                                                           | Chartres 2020                                       | Knowledge & Skills |

|                                                                      |                    |                                                                 |                                                                                                                                                                                           |                                                                                                                                                                                                                                                                                                                                                                                                                                                          |                                                                       |                                     |
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| <b>13. Developing Recommendations and Determining their Strength</b> | All                | Patients & caregivers<br>Providers of care                      | 13.1 Guideline developers should provide advance preparation materials and resources prior to meetings and throughout the project.                                                        | “When recruiting patients and consumers, guideline developers should establish the purpose of their engagement and provide preparatory materials and a point of contact.” (Armstrong 2017)                                                                                                                                                                                                                                                               | Armstrong 2017; Brouwers 2018; Chalmers 2017; Jarret 2004; Lanza 2000 | Environmental context and resources |
|                                                                      | Patients<br>Public | Patients<br>Public<br>Providers of care<br>Guideline developers | 13.2 Guideline developers should offer explanations and promote the use of plain language to ensure that participation in the guideline development process is accessible to all members. | “Training all committee members, including health professionals to communicate in plain language, will serve to further include lay members in a meaningful manner.” (Biggane 2022)<br><br>“A recurrent notion was that of professionals speaking a “foreign language” – using jargon, which was learnable for some and frustrating for others. A pivotal factor seemed to be the extent to which people explained terms to one another.” (Harding 2010) | Biggane 2022; Chalmers 2017; Harding 2010; Piggott 2021               | Knowledge                           |
| <b>14. Wording of Recommendations and of Considerations of</b>       | All                | Patients<br>Guideline developers                                | 14.1 Guideline developers should consider using online                                                                                                                                    | “Online modified-Delphi processes can be used to identify consensus on which review summaries                                                                                                                                                                                                                                                                                                                                                            | Pottie 2021, Khodyakov 2019/Grant 2021, Lamontagne 2021               | Other                               |

|                                               |                                                                                  |                      |                                                                                                                                                |                                                                                                                                                                                                                                                        |                 |               |
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| <b>Implementation, Feasibility and Equity</b> |                                                                                  |                      | methods/tools to refine recommendations.                                                                                                       | and conclusions patients find most believable, credible, or useful. Guideline development groups can then engage patients in recommendation development to ensure that recommendations facilitate patient-centered care.” (Grant 2021)                 |                 |               |
| <b>15. Reporting and Peer Review</b>          | Patients<br>Public<br>Providers of care<br>Product makers<br>Peer review editors | Guideline developers | 15.1 Stakeholder engagement in the external review process may benefit from longer review periods combined with individualised reminder emails | “Other measures to encourage people to take part in the external review of dermatology guidelines might include longer review periods combined with individualised reminder emails to people within all categories of stakeholders.” (Bartelmann 2020) | Bartelmann 2020 | Reinforcement |
| <b>16. Dissemination and Implementation</b>   | Patients                                                                         | Guideline developers | 16.1 Online delphi methods can be used to engage patients in identifying implementation considerations and dissemination strategies.           | “Once CPGs have been finalized, patients can then assist in disseminating and implementing CPG recommendations, such as facilitating the use of acceptable terminology in CPG recommendations and lay language summaries. For example,                 | Grant 2021      | Other         |

|                               |          |                      |                                                                                                                                                                  |                                                                                                                                                                                                                                                                                                                                                                                       |            |       |
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|                               |          |                      |                                                                                                                                                                  | patients and patient advocacy groups can endorse guidelines to improve their legitimacy and trustworthiness to patient communities, as well as patient adherence to guideline recommendations. Online modified-Delphi processes can be used to engage patients in identifying items to include in decision aids, as well as barriers and solutions to guideline uptake.” (Grant 2021) |            |       |
| <b>17. Evaluation and Use</b> | -        |                      | None reported                                                                                                                                                    |                                                                                                                                                                                                                                                                                                                                                                                       | -          | -     |
| <b>18. Updating</b>           | Patients | Guideline developers | 18.1 Using an online Delphi process to assess the patient-centeredness of guideline recommendations may help facilitate patient engagement in guideline updates. | “Lastly, patients can assist in updating and evaluating CPGs, such as determining if and when guidelines require an update, assessing whether patients made meaningful contributions to a CPG, and informing improvements to future patient engagement strategies in CPG development. To facilitate patient engagement at this stage, online modified-                                | Grant 2021 | Other |

|  |  |  |  |                                                                                                                                                                                                                                                                                                                                                      |  |  |
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|  |  |  |  | <p>Delphi processes can be used to continually assess the current patient-centeredness of CPG recommendations. For example, patients can define and prioritize significant changes in views from the patient community since a CPG has been developed, such as outcomes and interventions that should be included in a CPG update.” (Grant 2021)</p> |  |  |
|--|--|--|--|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--|--|

## 1. Organization, Budget, Planning and Training

### BARRIERS

**1.1 A lack of human and financial resources at the organisational level limited the collaborative development of recommendations.** Resource constraints were a general concern across several studies, which acted as a barrier to engaging patients, caregivers, patient organisations, and providers of care. Three studies specifically recognized the high level of human resources necessary to coordinate multi-stakeholder engagement efforts (Den Breejen 2014; Ilott 2006; van Wersch 2001), citing the need for dedicated paid staff or coordinator positions to handle the logistics of consultations, which was particularly challenging for smaller organisations (Ilott 2006). Specific engagement methods, such as surveys, interviews or focus groups, were identified as being desirable to capture the patient perspective in guidelines, but not all organisations would have the resources for such work (Hämeen-Anttila 2016).

**1.2 Guideline development organisations have unclear goals related to patient and public engagement.** Guideline developers and organisations often lack clear goals about patient and public engagement, being described as “largely implicit or articulated in vague terms [...] which makes it difficult to assess their success or failure” (Boivin 2010). Patients who are engaged in guideline development may have different understandings of the goal of their involvement; in one study patients believed that their involvement was a mechanism to change their own healthcare and disclose personal health problems, which derailed the guideline development group from its true purpose (Lanza 2000).

**1.3 Guideline developers deliberately exclude patients from guideline development.** Guideline developers may choose not to engage patients because they feel like the clinical practice guideline topic is too complex to require patient input (van de Bovenkamp 2015) or because they feel that patient contributions are too general for the question the specific guideline is trying to answer (Brouwers 2018). Patient organisations do not always see the added value of participation either, as they may think that professionals already have the knowledge needed to develop a good guideline, and patients provide little added-value (van de Bovenkamp 2015). In one study, guideline developers intentionally had no formal patient or public involvement to prevent tokenism (Selby 2017).



## **FACILITATORS**

***1.1 Guideline development organisations should implement formal stakeholder engagement models into their everyday operations.*** To facilitate the engagement of providers of care, patients and caregivers, several professional bodies and organisations have invested considerable resources in stakeholder relations. For example, incorporating stakeholding activities into their business plan and including these in the job description of professional guideline development coordinators (Ilott 2006). This can facilitate engagement as the stakeholder engagement process becomes a part of the organisation's everyday work. Guideline developers may also develop and incorporate engagement models into their guideline development (organisational) processes for future use (Ilott 2006; van der Ham 2014). This model sometimes results from patient and stakeholder engagement being a criterion to get development grants (van de Bovenkamp 2015). In the published literature, clear, effective organisational leadership was an important enabler for stakeholder-engaged work (Walker 2022). Two engagement models have been evaluated: The Heart and Stroke Foundation of Canada developed an inclusive and integrated model of patient engagement named the Community Consultation and Review Panel (CCRP). The goal of the CCRP model was to facilitate development of recommendations that are grounded in real-life experience and the unique needs of patients while maintaining scientific rigour. The model provided members with opportunities to regularly review and offer feedback on draft recommendations from the scientific writing group, and provided context related to their values and preferences (Lindsay 2020). Similarly, a team of researchers from RAND and clinicians, caregivers, and patients from the Parent Project Muscular Dystrophy (PPMD) developed the RAND/ PPMD Patient-Centeredness Method (RPM). The RPM is an online modified-Delphi approach to patient engagement that allows patients and their representatives to provide input on the patient centeredness of draft guideline recommendations (Armstrong 2019). This model has been suggested for use throughout all stages of guideline development to continuously engage a panel of patients and their representatives (Armstrong 2019).

***1.2 Provide stakeholders with financial compensation and/or professional accreditations.***

Guideline development organisations may facilitate stakeholder involvement by budgeting for and obtaining funds to financially compensate stakeholders, particularly for patient representatives as well as caregivers, patient organisations, and providers of care. This may

include reimbursement for travel, printing, and other costs (e.g. childcare), as well as honoraria (financial support) to cover their time (Armstrong 2017; Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Iltott 2006; Jarret 2004). For providers of care, alternative compensation in the form of professional accreditations (e.g. CME) could be considered (Iltott 2006). For all stakeholders, inclusion in publications if they meet the criteria for authorship should also be considered. One study suggested that covering expenses and providing compensation can facilitate engagement of patients/public from diverse identities (e.g. from different geographies, race/ethnicity, culture, language, etc) (Synnot 2022).

### ***1.3 Guideline developers should provide training for patient and public stakeholders.***

A recurring theme was the need for training or education and support for all stakeholders, but particularly patient and public members, to facilitate better understanding of the guideline development process (Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Hämeen-Anttila 2016; Jarret 2004; van de Bovenkamp 2015). Training on how to effectively participate could be provided before and during involvement. A study by Daraz et al. (2019) found that pre-engagement training may be especially beneficial for patients as this can help assist with incorporating their input and preferences early into the guideline development process. They recommended that the training should be delivered prior to the first guideline panel meeting and before the final recommendation deliberation (Daraz 2019). Training could include information about clinical guidelines, research methodology, evidence frameworks, and basic statistics. Daraz 2019 described a training curriculum targeting patients' knowledge, skills, and attitudes based on content from the Grading of Recommendations Assessment, Development and Evaluation (GRADE) Working Group and readability principles. In a webinar, the focus was on didactics and concepts, and in a workshop, the focus was on roleplay and practical issues (Daraz 2019). Other literature suggests that training could come in the form of orientation sessions by teleconference or in-person, by watching a video of a guideline development group in action, or through mentoring opportunities from other patient/public representatives. Patient and public members should also be advised of various resources and support available and be provided with examples illustrating the impact of lay members in previous guideline developments (Biggane 2022). Guideline development group members should be notified of the level of training and resources that patients receive before meetings. A study by Chartres 2020 suggested that training all guideline development group members who are unfamiliar with

evidence frameworks should be an ongoing process, and Armstrong 2020 suggests this promotes co-learning.

## **2. Priority setting**

We did not identify any barriers and facilitators to stakeholder engagement specific to the priority setting stage of guideline development.

## **3. Guideline group membership**

### **BARRIERS**

#### ***3.1 Challenges identifying “representative” patients to engage in guideline development.***

Several studies reported that identifying patients, caregivers and patient representatives to participate in clinical practice guideline development is a challenge (Biggane 2022; Kim 2021). There is some debate as to whether patients involved in guideline development should represent the larger patient population or simply provide their own unique patient perspective. In several studies, guideline development groups were uncertain about whether patients were being properly represented (Brouwers 2018; Chalmers 2017; Hämeen-Anttila 2016; Lanza 2000; van der Ham 2014). One article specifically expressed concerns regarding patient proxies (such as caregivers or healthcare professionals) who may be included within a guideline development group to represent the patient’s interests, stating “interests and needs of patients [are] sometimes distinct to those of [other stakeholders]” (Biggane 2022). Guideline development group members pointed out that it may be difficult to find appropriate persons from the target group who would be capable of representing the larger patient population. Consequently, balancing representation and stakeholder opinion was difficult in participatory development processes (Walker 2022). In two studies, it was difficult to represent the diversity of the target group in the guideline development process with respect to ethnic diversity and health conditions (Kim 2021; van der Ham 2014). Armstrong 2020 found that the experiences and perspectives of patients included in guideline development may be different from those who choose not to participate in the guideline development process, and emphasised that self- selection may occur and that diverse patient perspectives may be insufficiently articulated.

#### ***3.2 Patients perceive ‘tokenistic engagement’ which limits their ability to meaningfully***

***contribute to guideline development.*** Seven articles identified tokenistic engagement, that is, offering engagement as a gesture, as a challenge to meaningfully engaging patients (Armstrong 2017; Atkins 2013; Biggane 2022; Brouwers 2017; Daraz 2019; Jarret 2004). Instead of being equal members of guideline development groups, some patients expressed frustration and felt they were included merely to fill a quota (Biggane 2022). This was described as being a “tokenized member of the team” rather than a fully integrated member (Brouwers 2017) and highlighted the need for strategies to guard against tokenism, ensure that all members contributed equally to discussion and decision making, and ensure that the patient voice was adequately captured in the final guideline (Armstrong 2017; Atkins 2013).

***3.3 Language fluency may limit the representativeness of patients and their ability to engage in group processes.*** Three studies highlighted the potential for language and manner of speaking to become barriers to patient engagement (Armstrong 2017; Chalmers 2017; Piggott 2021). For example, there may be a requirement for patients to speak only English in order to participate in guideline development meetings, leading to lack of representation from other linguistic groups (Chalmers 2017). Community members (patients and the public) also expressed concern that physicians from other countries who are members of the guideline development group may have accents or speak differently, making it difficult for them to understand discussion (Armstrong 2017). Stakeholders [guideline methodologists and content experts] have reported that different cultures and languages can have a major impact on group processes and that respect and consideration for this should be given by all guideline development group members (Piggott 2021).

## **FACILITATORS**

***3.1 Patient/public and wider stakeholder selection should consider the representativeness and diversity of membership.*** Several studies mentioned the importance of recruiting ‘appropriate’ or ‘representative’ patients to participate in guideline development (Armstrong 2017; Boivin 2010; Brouwers 2018; Chalmers 2017; Den Breejen 2014; Harding 2010; van der Ham 2014; van Wersch 2001). Clear eligibility criteria is a facilitator to engaging people who represent the types of lived experience that are sought by the guideline developers; a “job description” may help to specify the knowledge, experience and social skills needed. Online modified-Delphi processes could be used to propose and prioritise desired characteristics of guideline development group

members, including the type of expertise or experience group members should have (Armstrong 2019). Involving multiple patient representatives or patient advocates may facilitate peer support and representation.

Project leaders may consider diversity as they are recruiting - this includes perspectives, roles, ethnicities, and other differences within the patient group. In order to properly consult marginalised patient sub-groups, it may be necessary to pay special attention to their inclusion. A study by Armstrong 2020 found that local organisations could help to identify relevant patient representatives, as this could aid in overcoming recruitment difficulties as well as limit the use of resources required for their engagement. Using this recruitment approach, patient stakeholders had at least a basic knowledge of the relevant medical terminology (Armstrong 2020). An additional article emphasised the importance of establishing respectful partnerships with organisations that work with or represent diverse groups, as they can assist with recruitment (Synnot 2022). Power imbalances between members (e.g. between content experts and evidence review groups) could also potentially be minimised by paying attention to differences in age, experience, gender, or region and intentionally selecting members with diverse experiences and characteristics (Chartres 2020). One study highlighted that special considerations would be needed to engage children in guideline development, as “clear requirements to give children a voice in the process of guideline development do not exist” (Schalkers 2017).

***3.2 The recruitment of patient representatives should prioritise the identification of knowledgeable and experienced patient representatives.*** Many articles suggested that guideline development groups should recruit patients that are familiar with the guideline topic and process (Armstrong 2017; Biggane 2022; Brouwers 2018; Hämeen-Anttila 2016; Kim 2021; van der Ham 2014). Patient panellists should be knowledgeable, have relevant expertise, and be willing to actively contribute and ask questions (Armstrong 2017). Several potential groups were identified – such as patient trainers, peer support patients, and patient representatives in hospital ethics committees – that could be engaged without extra training because they are already experienced in illustrating patient views (Armstrong 2017). One study shared the perspective of a ‘professional’ patient representative, whose professional role provided additional insight and enabled her to share knowledge developed through her work. This professional background and role enabled her to be involved throughout the clinical guideline development process, in a way that was different from other patient and public members. (Biggane 2022). Patient advocates,

who had experience sharing their opinions in multidisciplinary groups and committees, felt more confident in guideline development groups. The ‘helicopter view’ that certain patient representatives have is considered beneficial, meaning that they have an overview of a wide range of experiences from a range of service users (Armstrong 2017; Brouwers 2018; Hämeen-Anttila 2016; van der Ham 2014). One study explicitly stated that guideline developers should aim to involve patients with prior engagement or guideline experience (Kim 2021). In another study, patient/public members representing patient charities required less training in order to be involved due to their professional background and role (Biggane 2022). Whilst these members did sometimes have difficulty understanding their role and the process of guideline development, when compared to patient members with lived experience of a condition, members from patient charities did not seem to struggle with the technical content of the guideline development. These “professional” patient/public members were often able to understand the scientific content and language they were presented with during the guideline development process (Biggane 2022).

**3.3 Guideline developers should consider the ratio and diversity characteristics of patients and physicians within a guideline panel.** In one article (Armstrong 2017), some patients felt that they would be comfortable as the only patient representative on a guideline panel, whereas others said this would make them feel like they didn’t belong and it would be intimidating. Those who expressed feeling more comfortable with the idea of serving as the sole patient representative were older and described more experience with either physicians or group dynamics. There was general consensus that including multiple patient representatives would be ideal. Some patients indicated that simply having multiple patient members would be enough for them to feel comfortable, while others indicated that they would want there to be an equal number of patients and physicians. Regardless of panel composition ratio, patients agreed that smaller panels would make them more comfortable presenting their views, particularly to physician panellists (Armstrong 2017). In this study, most patients said that physician race or ethnicity would not matter, but several African-American participants indicated that they would be more comfortable if there was a physician of the same race on the panel.

**3.4 Guideline development benefits from the inclusion of methodological experts.** Three studies identified the engagement of principal investigators as an enabler to developing guidelines (Chartres 2020; Kim 2021; Piggott 2021). Specifically, they recommended including experts in qualitative research and systematic reviews to offset the training requirements needed by other

guideline group members (Kim 2021). In two studies, these “technical experts” or “methodologists” were described as working as part of an evidence review group, independent from the guideline development group (Chartres 2020; Piggott 2021). However, their inclusion in the larger guideline development group was identified as an important enabler for guideline development (Chartres 2020; Kim 2021; Piggott 2021). Methodologists who are members of guideline development groups could facilitate discussions of the reviews presented by the evidence review groups. Their understanding of the challenges associated with evidence review could bridge a gap in communication between the guideline development group and evidence review groups (Chartres 2020). By integrating the groups, with subgroups to evaluate the evidence for particular questions, the tensions from lack of contact and communication may be significantly reduced (Chartres 2020).

***3.5 Taking a network approach to guideline development benefits the engagement of stakeholders in the guideline development process.*** Studies suggested that a network approach (e.g. formation of a network of organisations or individuals in guideline development) could facilitate the engagement of patients, providers of care, payers of health services/research, policymakers and principal investigators. Guideline development groups can use these networks to identify potential members and/or to share learning, operational systems, and resources among organisations. In order to encourage follow-up, guideline development groups should decide on the communications mechanisms that they will use to maintain their network - emails, newsletters, and journals were common choices. Updates are important to maintain cohesion and can be done through email or webcast with select groups of stakeholders (Carter 1995; Den Breejen 2014; Illott 2006; Jarret 2004; van der Ham 2014).

## **4. Establishing Guideline Group Processes**

### **BARRIERS**

***4.1 Unclear roles and responsibilities for guideline development group members may weaken or disrupt group processes.*** When organisations do not have clear stakeholder engagement goals, the precise roles and responsibilities of each member of a guideline development group is often unclear. In the literature, there was a lack of clear discussion about patients’, providers of care, and principal investigators’ roles and tasks in the guideline development groups, causing

representatives to be uncertain as to what was expected of them. Patients, in particular, were described as not being prepared for their role and were not aware of how their input influenced the guideline (Kim 2021). This can lead to members misinterpreting why they were selected and what they are supposed to do in the group, leading to tension between stakeholder groups (Chartres 2020), creating confusion over how to use resources and which groups to involve (Carter 1995; Jarret 2004; Lanza 2000; van der Ham 2014).

**4.2 Failing to manage power imbalances between patients and providers of care may affect group dynamics and limit patient engagement.** Power differences between professionals and patients may cause issues within the guideline development group (Armstrong 2017; Atkins 2013; Carter 1995; Harding 2010; Kredo 2018). If some personalities in the group are stronger than others, especially if members are posturing or power positioning, it may influence how patients participate in discussions. Guideline development meetings might be dominated by some stakeholders to the exclusion of others. Perceived power imbalances can come from various sources - patients may not be comfortable if their own physician is on the panel (Armstrong 2017), patients may interpret the use and explanation of jargon by other stakeholders as a display of power, and discussion chairs or leaders may not consider patient input. This can also extend to the national level, where territoriality and “turf protection” can interfere with national coordination and development efforts (Carter 1995).

**4.3 A lack of confidence in their skills and legitimacy may reduce patient members’ level of engagement.** Lack of confidence was a commonly cited barrier to patients sharing their opinions in guideline development groups, especially if they did not have prior experience participating in guideline development. Some patients did not want to appear uninformed or unhelpful when talking with experts and felt that their experiences were not as important as professional opinion (Armstrong 2017; Brouwers 2018; Chalmers 2017; Harding 2010). This led to feelings of nervousness among patients and led to difficulties speaking up and voicing their opinions (Kim 2021).

## **FACILITATORS**

**4.1 Clear roles and expectations for stakeholder involvement in guideline development facilitate meaningful engagement, particularly for patients and principal investigators.** All



stakeholders in a guideline development group should be aware of their roles, the tasks to be completed, and the timelines within which the guidelines are expected to be completed (Biggane 2022; Piggott 2021). In the literature, the most common role in need of more clarity was the role of patients. Patient roles should be clarified so that they understand what is expected of them, as well as the roles of others in the group. This can be done through orientation meetings delivered by a coordinator/navigator where patients can be told about and ask questions about the purpose and limits of their role. To clarify overall participant roles, guideline group members should have a strong understanding of their own and others' experiences, clear project objectives and principles (Boivin 2010; Brouwers 2017; Brouwers 2018; Carter 1995; Chalmers 2017; Den Breejen 2014; Duff 1996; Ilott 2006; Lanza 2000). One article highlighted that the role of the evidence review groups needs to be clearly articulated from the start and reinforced throughout the guideline development process (Chartres 2020). The evidence review groups also need to feel appropriately supported in their roles when presenting the evidence reviews, particularly by the working groups chairs (Chartres 2020).

#### ***4.2 Guideline developers should foster group processes that promote patient empowerment.***

Empowering and fostering self-determination in patients can facilitate their participation in guideline development and can promote the involvement of patients as equal members of guideline panels. Personal motivators that empowered patient engagement included a feeling of appreciation from the members of the guideline development group, acknowledging patient effort, providing information and reading material in advance, and/or pre-specifying mechanisms for soliciting opinions. Panel chairs can also give the option of passing on questions or submitting written answers if patients feel uncomfortable in discussions, as well as creating separate spaces for patient feedback. Protected opportunities to share patient feedback, such as dedicated slots on meeting agendas, also helped patients to feel more capable of contributing. Two studies reported that patient-specific training resulted in feelings of empowerment (Biggane 2022) and improved self-efficacy and confidence (Daraz 2019). During training, patients developed a script to use during panel deliberations (e.g., what to do when the conversation includes too much jargon) and an instruction sheet for other panellists on how to empower and engage patients (Daraz 2019) (Armstrong 2017; Armstrong 2019; Biggane 2022; Brouwers 2017; Chalmers 2017; Ilott 2006; Jarret 2004; van Wersch 2001).

#### ***4.3 Allowing stakeholders to identify their preferred mode of participation can facilitate more***

***equitable and accessible engagement.*** There are often individual preferences around engagement, and guideline development stakeholders should be asked about these preferences. Guideline developers can consider engaging all stakeholders in-person and virtually. For providers of healthcare, there is a preference to network, collaborate and provide feedback in-person rather than online and to participate in structured discussions (Selby 2017). Direct, in-person engagement can also encourage patients to participate. This may include holding structured workshops in-person whenever possible as well as prompting patients during discussions. In-person meetings should be held at venues which are physically accessible. Where travel is prevented through ill health or other issues, teleconference facilities should be offered to avoid excluding patients' valuable contributions (Armstrong 2017; Chalmers 2017; Selby 2017). Patients and caregivers also reported online engagement processes to be a convenient way to overcome physical, geographical, and time constraints (Armstrong 2019; Brouwers 2018). However, online methods can preference the participation of well-educated, online-proficient consumers with internet access and this should be taken into consideration (Synnot 2022). Online methods can be cost-effective ways to engage the public and patient groups (Bartelmann 2020). In one study of the RAND/PPMD Patient-Centeredness Method (RPM), the convenience of an online approach was amplified by the asynchronous nature of the engagement process. Participants found the engagement process accommodating given that they could complete each round at their convenience rather than finishing all of it in one sitting (Armstrong 2019). Online methods facilitate engagement for people with physical or social disabilities and people who are disadvantaged and living in low and low-middle income countries, (Synnot 2022). A study among people with disability (traumatic brain injury) found that focus group discussions and online Wiki's were acceptable and feasible - with a slight preference for focus groups due to their social nature (Lamontagne 2021). However, in another study (Selby 2017), use of a wiki platform by providers of care was minimal (Selby 2017).

***4.4 Involving a skilled moderator/chair in the guideline development process can improve group processes and facilitate engagement.*** Effective guideline group process is impacted by the quality of organisation and support from guideline sponsor organisations and effective chairing (Piggott 2021). Chairs are essential to ensure that meetings occur according to an agenda and to provide clear expectations to ensure that discussions are held in a timely manner and activities are kept on target (Piggott 2021). In order to give patients and caregivers more

voice in guideline development groups, studies suggested appointing a skilled moderator with “a great deal of sensitivity” to lead the group (Armstrong 2017). Patient and caregiver members described a good chair as inclusive, skilled, open, honest, and able to influence guideline development group discussions (Jarret 2004), and able to actively support involvement of diverse groups (Synnot 2022). Two facilitation techniques were suggested for effectively engaging patients during meetings: notifying working group members of the training and resources that patients receive before meetings, and designating space for patient input in meeting agendas allowing patients to contextualise the clinical guideline content in a patient relevant manner (Biggane 2022). An effective chair can also help to enforce accessible discussions by asking patients for their opinions, asking professionals to explain their terminology, and protecting participant boundaries (Armstrong 2017; Brouwers 2018; Duff 1996; Harding 2010; Jarret 2004; Kim 2021). One study by Chartres 2020 found that power imbalances between working groups [providers of care] and evidence review groups [principal investigators] could be minimised through appropriate facilitation. Facilitation can allow groups to feel their opinions are respected and appreciated throughout the guideline development process.

***4.5 The provision of practical, technical and emotional support is a facilitator of engagement for all stakeholders.*** Support was identified as needed throughout the development process for all guideline development group members (Chartres 2020; Kim 2021). Guideline developers should designate an ongoing, proactive support person to provide practical, technical or emotional support (Synnot 2022). Providers of healthcare described their employers as fully supportive of them doing guideline development activities during work hours, and support took the form of both time and encouragement. Patients would benefit from support within their own patient organisations as well as collaborating centres. This can be either process-related support, such as monitoring of patients and their needs throughout the process by the project manager, or content related support, for example by organising collective input from patient organisations. Guideline development group leaders should also consider contracting with local agencies or consultants to support engagement in certain geographic areas (Den Breejen 2014; Duff 1996; Ilott 2006; Jarret 2004; Selby 2017; van der Ham 2014). Training all stakeholders in the skills required to take part in guideline development, such as communication and team-working, should also be provided (Kim 2021).

## 5. Identifying Target Audience and Topic Selection

### BARRIERS

We did not identify any evidence of barriers to stakeholder engagement specific to identifying a target audience or topic of guideline development.

### FACILITATORS

**5.1 Online Delphi methods facilitate patient engagement in topic selection and priority setting exercises.** One article described how patients can help nominate and prioritise topics that they find important (Armstrong 2019). Online modified-Delphi processes can be used by patients to systematically propose and develop consensus on the most important guideline topics. Specifically, patients could propose potential topics in an initial and open ended “brainstorming” round, and then prioritise them during subsequent rounds using close-ended rating or ranking questions. Alternatively, an online modified-Delphi process could help engage patients in prioritising an existing list of potential topics (Armstrong 2019).

## 6. Consumer and Stakeholder Involvement

### BARRIERS

**6.1 Patients’ engagement may be limited by feelings of intimidation derived from the process or other group members.** Intimidation may be a barrier to engaging patients. In one study, a few participants said that preferences are hard to accurately capture, that clinicians did not always accept patient preferences, and it was challenging to meaningfully involve patients (Kim 2021). Healthcare professional panel members are usually highly-trained and have experience working with other professionals but may not have experience working with those outside of their field and/or partnering with patients. Additionally, patient members may not have any training or experience within professional meetings, leading to hesitancy in commenting or questioning in the group discussions. This may be amplified if there is only one patient representative in the guideline development group. Some patients indicated that they did not want to appear uninformed or unhelpful, especially if the medical terminology and issues discussed are very complex. These participants may compare themselves to other guideline development group

members or have less influence, which can undermine confidence (Armstrong 2017; Atkins 2013; Brouwers 2017; Brouwers 2018; Duff 1996; Harding 2010; Jarret 2004).

## **FACILITATORS**

### ***6.1 Patients should be meaningfully engaged throughout the guideline development process.***

Involving patients in all different phases of the process instead of serving as occasional consultants makes their involvement more meaningful (Atkins 2013; Brouwers 2017; Brouwers 2018; Chalmers 2017; Jarret 2004; Selby 2017; van der Ham 2014). Guideline development groups need to support patients to enable them to meaningfully contribute during meetings. To avoid tokenism, patients should be involved as equal partners in decision-making processes. Leadership should ensure that these groups' opinions are incorporated into guideline development. Experienced members of the guideline development group could also help to facilitate meaningful patient engagement, especially if they have already interacted with medical professionals in a similar environment. Patients should be meaningfully involved right from the beginning stages of guideline development. This can include: inviting patients to speak during meetings, directly seeking their opinions during discussion, and endeavouring to ensure they understand the context and content of the development process (Biggane 2022).

## **7. Conflict of Interest Considerations**

## **BARRIERS**

***7.1 Patient engagement may be limited by concerns around confidentiality and judgement in the guideline development process.*** Some patients may refrain from sharing their opinion due to fears surrounding confidentiality or judgement (Armstrong 2017). Patients may be worried about privacy issues, stigma, and/or denial when sharing personal experiences. Patients may be under the care of a member of the guideline development group. This might not be a barrier for all patients, but it is possible that patients may feel coerced to participate or hide their true opinion if it is at odds with their physician's opinion (Armstrong 2017; Brouwers 2017; Chalmers 2017).

## **FACILITATORS**

**7.1 Guideline developers should manage conflicts of interest and patient concerns around confidentiality.** Project chairs should consider how they will manage patients' conflicts of interest, especially if patients are receiving care from another guideline group member. While patients who had good relationships with their physicians felt comfortable sharing their opinions, this is not always the case (Chalmers 2017). Chairs should take special care to maintain patient confidentiality and ensure that physicians do not disclose any personal patient information to the panel members without consent. Ideally, guideline developers should engage patients with no prior relationships with one another or the professionals on the panel (Armstrong 2017). One article suggested that an online modified-Delphi process could be used by patients to indicate which conflicts of interest and mitigation strategies are most salient for developing a given guideline (Armstrong 2019). In one study (Selby 2017), guideline developers created working groups and ensured that a majority of members were without conflicts of interest. Members of the guideline development group without commercial conflicts were asked to adjudicate the final recommendations, thereby diluting the effects of any potential commercial bias (Selby 2017).

## **8. (PICO) Question Generation**

### **BARRIERS**

**8.1 Patients and the public perceived that engagement in health outcome selection is outside of their purview.** In one study conducted in the UK, patient and public members became involved in other aspects of the clinical guideline development, but their involvement in selecting health outcomes was relatively limited (Biggane 2022). When asked, most patients and members of the public did not mention health outcomes as an area they were, or even wanted to be involved with. During committee meetings, they rarely participated in discussions about health outcomes or their selection and characterised this step of guideline development as a “system the technical team would go through”. How the role of patient/public members was perceived and the medical and scientific technicality of the guideline content all had a part in constraining their involvement in this topic (Biggane 2022).

**8.2 Time constraints and inflexible timelines may limit patient and public engagement in outcome selection and question generation.** Lack of time restricted the input of patient and

public members in health outcome selection (Biggane 2022) and patient and caregiver involvement in question generation (Köpke 2019). One study used surveys to gather PICO questions from an international sample of patients and caregivers, and reported that this was time and resource consuming (Köpke 2019). In the UK, this influenced guideline development later on, as the guideline development group was committed to the original questions and search terms developed earlier in the guideline process, and these were then not adaptable even if the resulting evidence was not relevant or understood by patient and public members (Biggane 2022).

## **FACILITATORS**

**8.1 Patient and public engagement may benefit from the provision of training specific to health outcome selection.** The NICE Public Involvement Programme (PIP) facilitates training sessions to introduce patient and public members to the guideline development process, including topics such as understanding scientific evidence, how it is used, and what are outcomes (Biggane 2022). Patient and public members largely described this training as “helpful” and “empowering”, praising the way their role, the process and the scientific terminology were explained (Biggane 2022). It is important that clinical guideline developers continue to understand and embrace the value of providing training opportunities to the various members of the committee. Training should include the importance of patient and public member involvement and the rationale for outcome selection. This includes training for: the chair of the committee, health professionals, technical team members and patient/public members themselves. Opportunities for this could include expanding on the outcome section of the PIP training session or communicating it at committee level in the early meetings (Biggane 2022).

**8.2 Guideline developers should consider establishing flexible processes and timelines.**

Guideline developers could explore whether there is flexibility in the timeline for the development of PICO questions and outcome selection. Specific suggestions include: seeking input to outcome selection at other junctures, establishing “break-out” working group about specific topics, and increasing the timeframe for guideline development by having pre-meetings or engagements with committee members (Biggane 2022).

**8.3 Guideline developers should use empirical methods for engaging with patients.** Once the guideline development group is established, patients can assist with developing the PICO

questions for the evidence review to ensure that its scope will be relevant and useful to real-world issues that the guideline aims to address. Online Delphi processes have been used to finalise the questions and outcomes. Specifically, patients can help refine clinical questions through an online modified-Delphi process that develops consensus on the importance of sub-populations, outcomes, aspects of treatments, and other elements (Armstrong 2019). One study suggested that this information could also be collected qualitatively using focus group discussions (Biggane 2022). They also recommended referring to already existing resources such as core outcome sets (Biggane 2022). A third study used an international survey to select PICO questions, which allowed participation of a geographically varied population and by being anonymous facilitated openness and trustworthiness (Köpke 2019). However, the authors noted that there was variable survey participation across countries, and that among those who did respond, they were online-proficient and well-educated participants, where the majority had completed post-secondary education (Köpke 2019).

**8.4 Guideline development should involve ‘Champions’ and representatives from disease-specific societies or organisations.** Disease-specific societies or advocacy groups can play a crucial role in the process of PICO question generation. In one study, the National Multiple Sclerosis (MS) Societies of participating European countries provided valuable feedback, particularly if stakeholders were both patients and MS Society members (Köpke 2019). For example, one specific task force member was an MS patient and a member of the MS International Federation, and of the Danish national MS society. She was key in bridging the issues of the MS patients to those of the health professionals and researchers (Köpke 2019), suggesting a valuable role of boundary spanners (i.e. institutions, groups, or individuals that straddle the divide between information producers and users) in health guideline development.

## **9. Considering Importance of Outcomes and Interventions, Values, Preferences and Utilities**

### **BARRIERS**

**9.1 Power imbalances between patients and providers of care in guideline development groups may promote unequal influence in decision-making.** There may be inequality in guideline development group members' influence on decision-making, especially if there is a perceived



hierarchy of opinion where the perspectives of patient representatives are considered to be less valuable than those of healthcare providers. Some guideline development group members may feel that patients do not easily fit into the decision-making structure. This can lead to limited influence of patient input, overturned group consensus, and a general undermining of the patient focus in guideline development. On the other hand, patients may not realise that their input is only one part of the guideline process and that the guideline development group is not obligated to follow the patients' recommendations. One study (Atkins 2013) reported that unequal influence was seen by some members as beneficial for decision making by preventing unfocussed discussion. (Atkins 2013; Harding 2010; Ilott 2006; Jarret 2004; Lanza 2000; van de Bovenkamp 2015).

## **FACILITATORS**

We did not identify any evidence of facilitators to engaging stakeholders in determining the importance of outcomes, interventions, values, preferences, and utilities.

## **10. Deciding what Evidence to Include and Searching for Evidence**

### **BARRIERS**

#### ***10.1 Patient engagement in guideline development may be limited by a lack of technical skills applicable to reviewing scientific evidence.***

Multiple studies mentioned the concern that the search, review, and discussion of scientific literature may be too technical or specialised for patient representatives. The average patient's lack of expertise - for both guideline development and literature reviews - was frequently cited as a barrier to engagement (Hämeen-Anttila 2016; Kredo 2018; Schalkers 2017; van de Bovenkamp 2015; van der Ham 2014)., for example: "He didn't have the sort of skills – his contribution could only be [...] from his experience that couldn't meaningfully be fed into that process" (Brouwers 2017).

#### ***10.2 The impact and depth of patient engagement may be limited by a devaluation of experiential knowledge.***

The strong focus on evidence-based medicine and the systems used to evaluate scientific studies and categorise them in terms of the strength of the evidence makes it hard to give the experiential knowledge of patients a place in the guideline as it does not fit this

categorization structure. One of the key challenges faced during guideline development processes is the dynamic tension between using an evidence-based approach (i.e., science, rigour and established expert contributions) versus tapping into tacit knowledge of all stakeholders (Walker 2022). Guideline development group members may have different ways of defining evidence, ranging from rigorous scientific trials to “evidence of experience”. Within this, the medical knowledge of professionals is considered superior to the experiential knowledge of patients (Atkins 2013; Harding 2010; van de Bovenkamp 2015), and that this might be even more the case with children than with adults, suggesting that children’s experience of illness is excluded from guideline development (Schalkers 2017). Several professionals indicated that it is often difficult to integrate input from patients into the guideline because it differed from other professional stakeholders (Armstrong 2020) or focused on aspects of care, such as the organisation of care or the patient-provider relationship, which are often not the focus of a guideline (van der Ham 2014). A single study also suggested that there are similar challenges engaging providers of healthcare and ensuring an appropriate balance between the evidence base, expert opinion and practitioner feedback (Selby 2017).

***10.3 Diverging disciplines and approaches between ‘evidence review groups’ (technical teams) and guideline development groups may promote intergroup conflict and division.*** Members of guideline development groups often have divergent disciplinary backgrounds (eg, physicians, toxicologists, clinical and public health researchers) compared to those on evidence review groups (eg, systematic review experts) and hold varying, sometimes conflicting beliefs about what constitutes ‘good’ evidence (Chartres 2020). In one study, content experts often viewed ‘good’ or ‘important’ evidence from the perspective of their own knowledge and expertise and not from the standards of methodological rigour used by the evidence review groups. These divergent roles and epistemological beliefs led to conflict and division between the groups. Many of the working committee members believed that in order to best protect the public’s health, the evidence presented by the review groups should at times be challenged (Chartres 2020).

## **FACILITATORS**

***10.1 Guideline developers should value experiential evidence and elicit patient knowledge beyond guideline development group membership.*** Patients are more likely to contribute if they

feel appreciated by other members of the guideline development group and/or that their input matters. There are opportunities to access patient input from other sources over and above patient membership in the guideline development group. Patient and caregiver organisations have good channels for gathering information from the patient perspective. One possible means of gathering patient views were surveys distributed by the patient organisations. Focus groups with patients and carers were also sources that guideline group members (including patient members) could draw on to inform their own contributions. Patients and caregivers appreciated interactive processes that include feedback loops (e.g. Delphi) and opportunities to directly engage with and learn from other patients. The iterative nature of data collection can foster better participant engagement by increasing their willingness to share and discuss their opinions and experiences with others, resulting in the development of new and shared perspectives (Armstrong 2017; Hämeen-Anttila 2016; Jarret 2004).

## **11. Summarizing Evidence and Considering Additional Information**

### **BARRIERS**

***11.1 Stakeholder engagement may be restricted by a separation of duties and limited communication between guideline development groups and evidence review groups.*** One study described how evidence review groups (composed primarily of researchers and methodologists) worked independently from the guideline development group for most of the process (Chartres 2020). The transfer of ideas, knowledge and expertise pertaining to the inclusion and presentation of evidence was limited by this separation. By not working with the evidence review groups regularly from the outset and understanding the methods thoroughly, some guideline development group members felt limited in their ability to contribute to the guideline process due to their lack of methodological knowledge or disagreement with the included evidence. The irregularity of the meetings at which the evidence reviews groups presented their findings to the guideline development group meant it became an ineffective way of communicating how the evidence was being evaluated (Chartres 2020). In turn, tensions developed between the two groups. Evidence review groups recognised an unequal and unfair power dynamic which led to them feeling that they were not valued or respected contributors to the guideline process (Chartres 2020). The evidence review groups members felt that the

tensions that were experienced could have been limited if guideline development groups were provided with clear information about the different roles of the two groups.

## **FACILITATORS**

We did not identify any evidence of facilitators to engaging stakeholders in summarising evidence and considering additional information.

### **12. Judging Quality, Strength or Certainty of a Body of Evidence**

#### **BARRIERS**

**12.1 Stakeholder engagement in evaluating evidence may be limited by an inadequate understanding of methods.** Guideline development group members and evidence review group members viewed understanding the methods [related to evaluating evidence] as a significant challenge to the current guideline development process. Inadequate understanding of the methods restricts the level of input certain guideline development group members can have in the process and creates tensions between the groups (Chartres 2020).

#### **FACILITATORS**

**12.1 Providing training on the frameworks employed in assessing the body of evidence may facilitate the engagement of guideline group members.** Pragmatic advice and training by the guideline development organisation for the guideline group members unfamiliar with the frameworks used to assess the body of evidence is necessary, not only at the commencement of the process but should be ongoing thereafter (Chartres 2020).

### **13. Developing Recommendations and Determining their Strength**

#### **BARRIERS**

**13.1 Patient and public engagement may be limited by their lack of content knowledge, clinical expertise and technical skills.** Some patients felt that they had a lack of clinical content

knowledge, insufficient academic education, guideline process training, and/or computer skills, which would hinder the complex scientific discussion and the recommendation development process. Unless they are educated about the guideline topic or have specific prior knowledge and training, patients may stay quiet during group discussion (Harding 2010; Schalkers 2017; Selby 2017; van de Bovenkamp 2015; van der Ham 2014). Additional barriers included the need for in-depth scientific discussion, which may not always be accessible for patients or caregivers, leading to the concern that discussions may be stifled if conversations are constantly being interrupted for lay explanations (Armstrong 2017; Armstrong 2020; Brouwers 2017; Brouwers 2018; Chalmers 2017; Den Breejen 2014). In one study, discussions among health professionals in committee meetings for this guideline indicated that they also believed that the content was too clinically driven for lay members to understand (Biggane 2022).

**13.2 Insufficient and competing demands of time may limit stakeholder engagement in recommendation development.** Time was a frequently cited barrier across multiple studies regarding the engagement of patients, caregivers, patient organisations, and providers of care. This included insufficient time for discussion and recommendation formulation, as well as the time-consuming nature of travel and attending meetings. Participants may feel rushed or unable to fulfil their responsibilities. For example, challenges were encountered in getting responses from professional societies within the four-week consultation deadlines, and these problems were more severe for organisations with limited capacity (under 2,000 members) (Ilott 2006). One study noted that it was difficult to keep patients interested in attending meetings because of conflicts with patients' work or personal lives (Brouwers 2018). A lack of dedicated time for guideline development may also lead to guideline development group members working voluntarily on their own time and afterhours (Armstrong 2017; Atkins 2013; Brouwers 2017; Brouwers 2018; Chalmers 2017; Den Breejen 2014; Ilott 2006; Kredo 2018). The length of time required to develop guidelines may also prevent healthcare professionals from engaging in their development (Bartelmann 2020). Clear timelines are also needed for the technical experts conducting evidence syntheses, so that sufficient time is allowed for the review of evidence in advance of guideline meetings (Piggott 2021). Patients should be given sufficient notice of meetings and allocated enough time to understand the materials and provide feedback (Brouwers 2018; Chalmers 2017; Duff 1996).

**13.3 Unequal access to information technology resources may limit stakeholders' ability to**

***engage in guideline development.*** Two studies, conducted among patients (Synnot 2022) and providers of care (Kredo 2018) identified that the guideline development process favoured those with access to and the ability to use the Internet and technology (Kredo 2018; Synnot 2022). One study noted that a patient representative had trouble carrying out her work because she had no printer and the documents she needed were soft copy only (Jarret 2004).

***13.4 Technical concepts and professional jargon may negatively impact patient and public comfortability and ability to contribute to the guideline development process.*** Patients thought meeting dialogue was sometimes overly technical and that professionals' use of jargon was like "speaking a foreign language" and represents a display of power (Armstrong 2020; Biggane 2022). This technical content comprised terminologies, abbreviations and topic content which lay members believed were largely only accessible to health professionals or those with specialised knowledge (Biggane 2022). Many patients may not be familiar with the medical language spoken (Armstrong 2020) and ask for clarification of terms and abbreviations. Patients without some research knowledge reported difficulties grasping the concept of the evidence framework and the statistics surrounding it. Some patients who found the documents and reading material too technical or scientific decided that they had no contribution to make on these issues and felt excluded. (Harding 2010; Jarret 2004; van Wersch 2001) Such patients were reported to be "participating observers of technical language to which they could hardly offer any input" (Brouwers 2018).

## **FACILITATORS**

***13.1 Guideline developers should provide advance preparation materials and resources prior to meetings and throughout the project.*** Providing resources to all stakeholders, especially hard-copy documents, before meetings and offering opportunities for clarification can help all guideline development group members contribute effectively. This may include supplying meeting agendas and talking points, questions, guidance and terms of reference documents, and/or materials to take notes with. There should also be a point of contact for guideline development groups members (Armstrong 2017); those in charge should make time to return calls and keep track of their contacts. It is particularly helpful for patients to meet with the chairs prior to meetings (and particularly the first meeting) so that they know what to expect and get to

know some other people in the meeting prior to joining larger group meetings (Armstrong 2017; Brouwers 2018; Chalmers 2017; Jarret 2004; Lanza 2000).

***13.2 Guideline developers should offer explanations and promote the use of plain language to ensure that participation in the guideline development process is accessible to all members.***

Guideline developers should ensure the language used in all aspects of the guideline development is accessible to all committee members (Biggane 2022). In particular technical experts must communicate concisely, effectively, and collaboratively toward the production of the guidelines (Piggott 2021), preferably in plain language avoiding scientific or medical abbreviations (Biggane 2022). Other activities may include translation of scientific literature and content into plain language formats as needed (Biggane 2022). A pivotal success factor for engaging patients seemed to be the extent to which people explained terms to one another. Project chairs or navigators can meet with patients prior to meetings so that patients can have an explanation of the context of the expected discussion. Guideline development group members can also use natural breaks in discussion to explain to patients in lay terms what is being discussed and ask patients specific questions where their input is needed. Some people linked this with an (often subtle) display of power which could be redistributed. Others felt these strategies contributed by preventing unhelpful assumptions from influencing proceedings (Chalmers 2017; Harding 2010).

**14. Wording of Recommendations and of Considerations of Implementation, Feasibility and Equity**

**BARRIERS**

We did not identify any evidence of barriers to stakeholder engagement specific to wording recommendations or considering implementation, feasibility and equity for guideline development.

**FACILITATORS**

***14.1 Guideline developers should consider using online methods/tools to refine recommendations.*** We identified 3 studies which suggested the use of online tools to engage

stakeholders in the refinement or wording of recommendations (Armstrong 2019; Lamontagne 2021; Pottie 2021). The GRADE-FACE is proposed as an efficient means to collect stakeholder perspectives on drafted recommendations whereby respondents assess the feasibility, acceptability, cost, and equity of each drafted recommendation on a GRADE rating scale and provide qualitative comments to justify and explain their response (Pottie 2021). The authors note that for underserved and disadvantaged populations, electronic surveys may not be feasible or appropriate. In-person semistructured interviews may be best suited to collect guideline perspectives from these groups (Pottie 2021). Similarly, the RPM is an online modified-Delphi approach to patient engagement in guideline development that allows patients and their representatives to provide input on the patient-centeredness of draft guideline recommendations by participating in an idea generational round, two rating rounds, and an online discussion round (Armstrong 2019). The RPM can also be used to explore the existence of consensus among key stakeholders on other dimensions of the Evidence-to-Decision framework, such as feasibility, equity, and resource use (Armstrong 2019), and issues related to disadvantaged patient subpopulations (Armstrong 2019). A third study proposed an online wiki approach (Lamontagne 2021). Participants considered the wiki method effective to the extent that it allowed for analysis and reflection before answering and because participants could express themselves without being stopped or interrupted. Other key advantages mentioned included anonymity, not feeling judged by others, the freedom to choose the time and place to answer the wiki questions, and doing it at the participant's own pace, respecting his or her capacity. However, participants were individuals with traumatic brain injury, and generally expressed preference for in-person options such as focus groups, as they more easily fit with the communication habits of individuals with a head injury who prefer oral expression to writing (Lamontagne 2021).

## **15. Reporting and Peer Review**

### **BARRIERS**

We did not identify any evidence of barriers to stakeholder engagement in the reporting and peer review stage of guideline development.



## **FACILITATORS**

***15.1 Stakeholder engagement in the external review process may benefit from longer review periods combined with individualised reminder emails.*** We identified only one study which reported facilitators to stakeholder engagement during the external peer review process of a guideline (Bartelmann 2020). They reported measures to encourage people to take part in the external review of guidelines might include longer review periods combined with individualised reminder emails to people within all categories of stakeholders. Paper-based advertisements in clinical practices and online advertising on patient-focused websites might be an option to identify patients and members of the general public who could participate. These measures, however, require time and funding that may not be available to the average guideline development group. Less expensive solutions might include the use of online platforms that allow external reviewers of guidelines to obtain recognition for their efforts even in cases where their reviews are anonymous. Having external review efforts formally recognised and rewarded by medical faculties or colleges might also have a salutary effect on participation rates (Bartelmann 2020).

## **16. Dissemination and Implementation**

### **BARRIERS**

We did not identify any evidence of barriers to stakeholder engagement specific to dissemination and implementation for guideline development.

## **FACILITATORS**

***16.1 Online delphi methods can be used to engage patients in identifying implementation considerations and dissemination strategies.*** Patients can assist in disseminating and implementing guideline recommendations. For example, patients and patient advocacy groups can endorse guidelines to improve their legitimacy and trustworthiness to patient communities, as well as increasing patient adherence to guideline recommendations. Online modified-Delphi

processes can be used to engage patients in identifying items to include in decision aids, as well as barriers and solutions to guideline uptake (Armstrong 2019).

## **17. Evaluation and Use**

We did not identify any evidence of barriers and facilitators to stakeholder engagement specific to evaluation and use during guideline development.

## **18. Updating**

### **BARRIERS**

***18.1 Guideline developers may face challenges engaging stakeholders in guideline updates due to insufficient financial resources.*** Two studies reported barriers to maintaining stakeholder engagement for patients, providers of care, policy makers, and principal investigators during guideline updates. In one study, lack of ongoing funding prevented the secretariat from keeping the original guideline development group engaged in guideline activities (Selby 2017). van der Ham 2014 also suggest that it can be difficult for patients to ‘catch up’ and meaningfully contribute if they were not involved in the original guideline.

### **FACILITATORS**

***18.1 Using an online Delphi process to assess the patient-centeredness of guideline recommendations may help facilitate patient engagement in guideline updates.*** According to one article, patients can assist in updating and evaluating guidelines, such as determining if and when guidelines require an update, assessing whether patients made meaningful contributions to a guideline, and informing improvements to future patient engagement strategies in guideline development. To facilitate patient engagement at this stage, online modified-Delphi processes can be used to continually assess the current patient centeredness of guideline recommendations. For example, patients can define and prioritise significant changes in views from the patient community since a guideline has been developed, such as outcomes and interventions that should be included in a guideline update (Armstrong 2019).

## 4.7 DISCUSSION

### Summary of main results

This qualitative evidence synthesis identified a total of 52 findings encompassing 24 barriers and 28 facilitators to stakeholder engagement in health guideline development. While the large majority of reported evidence is about patient engagement, many findings are applicable to all stakeholder groups and should be considered by guideline developers who aim to apply a multi-stakeholder engagement approach. In the table below, we summarise the barriers and their corresponding facilitators that are most strongly supported by literature and apply in a multi-stakeholder context.

**Table 7. Barriers and facilitators to multi-stakeholder engagement**

| Barriers                                                                           | Facilitators                                                                                                                                                                                                                                 |
|------------------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Lack of organisational resources to support engagement activities.                 | Implementation of an organisational stakeholder engagement model, including financial resources to compensate stakeholders and sustain engagement activities.                                                                                |
| Selection of “representative” stakeholders.                                        | Recruit knowledgeable and experienced stakeholders, with a focus on diverse perspectives, roles, and personal characteristics. Consider a network approach to identify appropriate and committed stakeholders.                               |
| Unclear roles and responsibilities.                                                | Clarify stakeholder roles from the beginning and reinforce throughout the guideline development process.                                                                                                                                     |
| Tokenism.                                                                          | Promote the meaningful engagement of stakeholders by providing practical, technical and emotional support. Engagement throughout all stages of guideline development supports more meaningful engagement compared to occasional involvement. |
| Power imbalances and unequal influence on decision-making.                         | Enlist a skilled moderator/chair to manage group dynamics. Offer explanations and promote plain language as-needed.                                                                                                                          |
| Lack of technical skills and content knowledge to support evidence-based medicine. | Provide training to empower stakeholders and improve self-efficacy and confidence in guideline development skills. Training should focus on evidence-based methods such as systematic reviews and evidence-to-decision frameworks.           |

|                   |                                                                                                                               |
|-------------------|-------------------------------------------------------------------------------------------------------------------------------|
| Insufficient time | Establish flexible processes and timelines. Consider stakeholders preferences for in-person and online engagement activities. |
|-------------------|-------------------------------------------------------------------------------------------------------------------------------|

## Overall Completeness and Applicability of Evidence

The evidence identified in this systematic review covers many topics of guideline development, however we did not identify any barriers or facilitators for some critical topics. Notably, we did not identify any evidence of barriers or facilitators for stakeholder engagement in priority setting, a surprising finding given the promotion of stakeholder engagement in this area (e.g., Grill 2021; Manafò 2018) and the international recognition of the James Lind Alliance approach (James Lind Alliance, n.d.). Additionally, evidence was also lacking for the following topics, and these remain areas in need of further research:

- Identifying Target Audience and Topic Selection
- Considering Importance of Outcomes and Interventions, Values, Preferences and Utilities
- Summarising Evidence and Considering Additional Information
- Wording of Recommendations and of Considerations of Implementation, Feasibility and Equity
- Reporting and Peer Review
- Dissemination and Implementation
- Evaluation and Use

Additionally, the availability of evidence was not evenly distributed across stakeholder groups. Patients and caregivers, members of the public, providers of care and principal investigators were most commonly engaged in guideline activities, and several barriers and facilitators to their engagement were identified. Other groups, such as policymakers, program managers, purchasers, peer review journal editors, product makers, and payers of research were infrequently engaged in guideline activities, and little to no barriers and facilitators to their engagement were reported. Despite this, many of the barriers and facilitators reported would apply to all stakeholder groups within the context of guideline development. To address this gap in the literature, additional research is being undertaken by the MuSE Consortium to establish stakeholder preferences for

engagement in guideline development (Magwood (submitted); Petkovic (submitted #2); Petkovic (submitted); Petkovic 2020).

The findings of this review have widespread applicability to any organisation or government that produces health recommendations or guidelines, including health technology assessments. However, the findings should be considered with some reservations. Almost all evidence in this review originates from high-income settings and is conducted among privileged and well-educated participants with good access to resources and technology. Very little information was available from equity-deserving groups and low-income settings. It remains unclear whether there are particular conditions or guideline settings (e.g., public health vs health system) that influence the engagement of diverse stakeholders. Country-level capabilities to form national guideline groups are not the same across the globe (for e.g., we do not see national guidelines from all LMIC countries). This could be because of their limited experience in framing guidelines and recommendations, which needs to be considered when international guidelines are prepared. To decolonise the guideline development process, guideline development groups should be balanced between representatives from global south and global north countries. Novel methods to include the voice of patients who have experienced poverty-related diseases should be developed to establish equity in guideline development.

### **Quality of the evidence**

The quality of the available evidence varies, with the best evidence found for patients and the public. This is because these stakeholder groups were often the focus of the included studies, which supports the growing movement for co-production in health. Studies involving patients and the public were more frequently prospective, included ethical approval, and employed rigorous methods such as ethnographic approaches and cross-over studies. For other stakeholder groups, studies were often retrospective and descriptive. Our CASP critical appraisal indicates that the most common concern regarding study quality was a lack of reflexivity/positionality by the study authors and a focus on the perspectives of the guideline developers rather than the stakeholders themselves. More high-quality prospective research is needed to capture the perspectives and preferences of multiple stakeholder groups.

Additionally, stakeholder characteristics were infrequently reported, and rarely available according to the PROGRESS-Plus acronym. This limited our ability to make conclusions about stakeholder engagement across diverse equity characteristics and settings. Ongoing updates to reporting guidance, such as the CONSORT-Equity 2017 extension for better reporting of health equity in randomised trials (Welch 2017) and the STROBE-equity reporting guideline to improve social justice in observational studies (Funnel 2023), may help in this regard. This highlights a larger issue regarding the reporting of barriers and facilitators of guideline initiatives - while reporting guidance is available for particular research designs (e.g., PRISMA, CONSORT, STROBE) and guidelines generally (e.g., AGREE, RIGHT), none of these tools provide authors with assistance self-identifying challenges or lessons learned from their own work. Given this limitation, it is likely that barriers and facilitators exist beyond what is reported in the peer-reviewed literature.

### **Limitations and Potential Biases in the Review Process**

This qualitative evidence synthesis offers a comprehensive overview of the barriers and facilitators to stakeholder engagement across all topics of guideline development for 10 different stakeholder groups. However, a number of limitations should be noted: i) While we included diverse stakeholders in the development of the protocol to inform this review, no patients or members of the public were engaged in the development of the review findings. ii) While no barriers or facilitators were identified for several topics of guideline development, it remains unknown where there are truly no issues, whether there were issues that were judged by authors as not worth commenting on in their publication, or whether study participants chose to remain silent about some topics. We did not attempt to contact study authors for missing data. iii) We included studies only if they were specific to at least one topic of guideline development. We excluded health technology assessments, as these processes will be covered in future work by the MuSE Consortium. iv) We searched for included studies in English language databases only, which may explain why the included studies originated primarily from high-income and anglophone countries. v) Finally, our review did not consider the role of “knowledge brokers” as a distinct interest-holder group. Knowledge brokers in health systems are important

intermediaries who play a critical role in bridging the gap between research, policy, and practice. A key component of the knowledge brokering process is facilitating collaboration and engagement among diverse stakeholders. Therefore, the inclusion of knowledge brokers may be an overlooked facilitator to engagement not considered in the current review, and this represents an important opportunity for future research.

### **Agreements and disagreements with other studies and reviews**

In the last decade, several reviews have acknowledged the importance of involving various stakeholders in the development of health guidelines (Armstrong & Bloom 2017; Bryant 2022; Cluzeau 2012; Kelson 2012; Légaré 2011; Pinho-Gomes 2022; Synnot 2022).

Particularly, the engagement of patients has become more common and the importance of engaging patients in the decision-making process has been recognized (Armstrong & Bloom 2017; Bryant 2022; Grant 2018; Kelson 2012; Légaré 2011; Pinho-Gomes 2022).

However, no previous reviews of the literature have considered the barriers and facilitators to engaging multiple stakeholder groups who are not traditionally a part of the guideline development process such as policymakers, program managers, payers of services or research, and peer review journal editors. Much of the existing literature focuses on the engagement of patients, the public, providers of care, and principal investigators.

One barrier that was consistent across our results and other reviews was challenges with guideline group membership. Our review found there can be challenges to recruit appropriate people from target groups that are capable of representing the wider patient population (Brouwers 2018; Chalmers 2017; Hämeen-Anttila 2016; Lanza 2000; van der Ham 2014).

There exists some debate as to whether patients engaged in guidelines should be “representative” -- there are no other group members who participate with the expectation of representation. For example, health care providers are diverse in the ways they practise and the contexts within which they practise, yet they are not expected to stand in for all providers. Additionally, there may be challenges in representing the target stakeholder group in terms of health conditions and ethnic diversity (van der Ham 2014). Similarly, a review by Légaré 2011 described challenges in ensuring appropriate representation of patients. In a review by Merner 2023, recruiting a range of

patients, caregivers and community members who were reflective of the clinic's demographic population was considered desirable, particularly by health providers.

Our review found that many studies emphasised the significance of choosing appropriate patients to be included in guideline development, with clear eligibility criteria in mind (Armstrong 2017; Boivin 2010; Brouwers 2018; Chalmers 2017; Den Breejen 2014; Harding 2010; van der Ham 2014; van Wersch 2001). Légaré 2011 suggested that participants should be as representative of the whole population as possible. Reviews by Légaré 2011 and Kelson 2012 found it can be challenging to identify appropriate patients to engage as stakeholders and identify stakeholders who are willing to contribute to guideline development. Recent publications have outlined factors for research teams to consider during identification and invitation of individual representatives in a multi-stakeholder partnership, including experience, inclusivity, willingness, communication skills, and time capacity (Parker 2022). A review by Grant 2018 concluded that as patients become more actively involved in clinical guideline development, additional research is needed for ways to engage large groups of diverse patients from various geographic locations. Using a “network approach” can help recruit representative and knowledgeable patients (or other stakeholders), however this risks recruiting the “usual suspects” and limiting perspectives.

Having a chair (moderator) that supports stakeholder-engaged guideline development was commonly noted as a facilitator. Our review found it is important to select a chair who can assist in asking patients for their opinions during discussions and offer explanation of terminology (Armstrong 2017; Brouwers 2018; Duff 1996; Harding 2010; Jarret 2004) and help with comprehension of discussion content (Chalmers 2017; Harding 2010). Légaré 2011 also emphasised the importance a chair has in supporting patients, ensuring patients are engaged and have a voice during discussions, and creating opportunities for meaningful contributions. The guideline chair can also ensure that all stakeholders are aware of their roles and responsibilities throughout guideline development. Our review found that all guideline members should clearly understand their roles and expectations within the group (Boivin 2010; Brouwers 2017; Brouwers 2018; Carter 1995; Chalmers 2017; Den Breejen 2014; Duff 1996; Ilott 2006; Lanza 2000). A review by Légaré 2011 supported this, stating every group member must be aware of their scope within the guideline development process with well-defined goals.



The devaluation of patient experiential knowledge was a similar barrier found in our review and others. van der Ham 2014 stated that there are challenges with including input from patients into guidelines because often their input focuses on aspects of care which may not be the focus of the guideline. Guidelines tend to focus on evidence-based medicine (i.e. empirical evidence) and this creates a challenge for including experiential knowledge of patients in guidelines (Atkins 2013; Harding 2010; Schalkers 2017; van de Bovenkamp 2015). A review by Pinho-Gomes 2022 found challenges and uncertainty around integration of patient experiences into guidelines that are heavily evidence-based. Reviews by Kelson 2012 and Légaré 2011 also found that a challenge exists around creating true evidence-based guidelines while including all relevant stakeholders, as the values and preferences of patients and caregivers may vary greatly from that of clinical experts. Armstrong & Bloom 2017 also found that differences between patient and physician views were significant barriers for the low frequency of public and patient engagement in guideline development. A review by Cluzeau 2012 also found that various stakeholders may hold different perspectives, thus placing differing weights on outcomes. It is because of these differences that guideline development groups should include the diverse perspectives of all stakeholders in decision-making (Cluzeau 2012).

Valuing patient contributions in the guideline development process was therefore identified as a facilitator. Our findings suggest that contributions from patients are to be equally valued, patients should have autonomy in the decision making processes, and should be able to freely provide feedback (Armstrong 2019; Chalmers 2017; Den Breejen 2014; Ilott 2006; Jarret 2004; van de Bovenkamp 2015). Similarly, a review by Kelson 2012 found that perspectives from both patients and caregivers are to be included and valued in the decision making processes for clinical guidelines. A review by Armstrong & Bloom 2017 also emphasised the importance that the public and patient groups have opportunities to contribute to guideline development. Pinho-Gomes 2022 found that the patient and public group are to be valued equally in guideline development, as they can provide unique and valuable insights. We found studies suggesting that patients are to be included in meetings when their input would be beneficial and relevant (Armstrong 2019; Chalmers 2017; Den Breejen 2014; Ilott 2006; Jarret 2004; van de Bovenkamp 2015).

Our review also found that involving patients at all stages of guideline development instead of serving occasionally provides more meaningful engagement (Atkins 2013; Brouwers 2017; Brouwers 2018; Chalmers 2017; Jarret 2004; Selby 2017; van der Ham 2014). Reviews by Légaré 2011 and Pinho-Gomes 2022 also emphasised that patients are to be involved at every stage of guideline development, particularly at the beginning. We found that patients were empowered when they were given the options and flexibility in communication, such as passing on questions or providing written answers if they were uncomfortable engaging in discussions (Armstrong 2017; Armstrong 2019; Brouwers 2017; Chalmers 2017; Ilott 2006; Jarret 2004; van Wersch 2001). Providing safe and confidential opportunities for patients to share feedback assisted patients to feel more qualified to contribute to guideline development (Armstrong 2017; Armstrong 2019; Brouwers 2017; Chalmers 2017; Ilott 2006; Jarret 2004; van Wersch 2001).

Additionally, our review found that guideline development processes favour stakeholders who have access to and know how to use technology and the Internet (Jarret 2004; Kredo 2018). Particularly, some patients may have insufficient computer skills needed to properly engage in the guideline development process (Harding 2010; Schalkers 2017; Selby 2017; van de Bovenkamp 2015; van der Ham 2014). Grant 2018 reported that some patients may not be able to participate using online methods due to lack of access to resources. Despite this, our review found that caregivers and patients preferred online engagement methods, as geographical and time constraints were removed (Armstrong 2019; Brouwers 2018). Some advantages of using online methods are that they provide opportunity for transparency in responses and allows representation of diverse patient perspectives (Grant 2018). Grant 2018 found online patient engagement was most successful when the guideline development group properly trains patients to ensure understanding of tasks. Two other reviews by Kelson 2012 and Légaré 2011 also emphasised the importance of having proper training and support for all to enhance collaborative work. Proper education, training, and support for all stakeholder groups helps to create a better understanding of the guideline development process (Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Hämeen-Anttila 2016; Jarret 2004; van de Bovenkamp 2015). This training can come in the form of online methods or in- person orientation sessions (Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Hämeen-Anttila 2016; Jarret

2004; van de Bovenkamp 2015).

Finally, our review found that resource constraints were barriers for engaging patients, caregivers, patient organisations and healthcare providers as there is a high level of coordination needed for stakeholder engagement (Brouwers 2017; Den Breejen 2014; Hämeen-Anttila 2016; Ilott 2006; van Wersch 2001). Reviews by Kelson 2012, Légaré 2011, and Pinho-Gomes 2022 also found that involving patients can be resource intensive. A review by Cluzeau 2012 mentioned that engaging and involving stakeholders can be costly for organisations with insufficient funds and may be a barrier in guideline development. Our review also found the time required for guideline development was a barrier because many stakeholders are required to work after hours on their own time while maintaining their own jobs (Armstrong 2017; Atkins 2013; Brouwers 2017; Brouwers 2018; Chalmers 2017; Den Breejen 2014; Ilott 2006; Kredo 2018). Reviews by Légaré 2011 and Pinho-Gomes 2022 also emphasised how certain stakeholders do not have time to engage in guideline development. It is not practical for members to take time off work and voluntarily contribute to guideline development (Légaré 2011). Stakeholders may be more engaged if there is financial compensation for involvement, for example covering travel and printing costs (Armstrong 2017; Boivin 2010; Brouwers 2017; Brouwers 2018; Chalmers 2017; Duff 1996; Ilott 2006; Jarret 2004). A review by Légaré 2011 also emphasised that financial reimbursement and assistance can help with stakeholder engagement. Financial aid can come in the form of covering travel expenses, loss of earnings, or provision of child care (Légaré 2011). Compensation can also address equity concerns by ensuring that underrepresented groups are not disadvantaged due to their inability to engage in guideline development without financial support. For example, providers of care may be able to contribute without financial compensation due to institutional support, but other stakeholders may rely on honoraria to cover costs associated with their participation. However, there is concern that financial compensation could introduce biases or conflicts of interest. Transparency around compensation is, therefore, key to maintaining trust in the process. Co-authorship of guideline publications also represents another form of acknowledgement which has traditionally been reserved for academic principal investigators or providers of care. However, there has been a shift toward recognizing the contributions of other stakeholders, such as patients, in guideline publications. Consequently, organizations must develop clear criteria for co-authorship that

reflect their different types of contributions to guideline development.

## **Authors' conclusions**

### **Implications for Practice and Policy**

This review identified barriers and facilitators to stakeholder engagement in health guideline development, allowing for the collation of current information on what works well in practice. The facilitators found will be used to develop an extension to the GIN-McMaster Checklist for Guideline Development and will support future efficient and productive multi-stakeholder work. We expect the findings to be considered by guideline development organisations such as GRADE, WHO, NICE and GIN. Notably, the majority of the current evidence focuses on patient engagement, and there are significant gaps in the literature which obscure or do not report on the engagement preferences of other stakeholder groups. Additionally, facilitators were not identified for all checklist topics so additional research should be done in order to understand how to best deal with known challenges. For example, how to manage diverging disciplinary perspectives, how to recruit stakeholders with diverse characteristics and experiences, or how to sustain stakeholder engagement to support future updates. Additionally, this work could be used as the basis for toolkits, template (e.g. role description) development and testing to offer guidance on 'maximising' stakeholder engagement. It argues for the development of training courses and tools to support meaningful involvement of all stakeholders, both public and professional. Taking a multi-stakeholder engagement approach to the development of health recommendations supports the growing movement of co-production in health care and policy, whereby patients/public are equal partners in shaping the healthcare system, including health guideline development (e.g. Batalden 2016).

### **Implications for Research**

The focus of this review was on barriers and facilitators to stakeholder engagement in the development of guidelines. As a follow up, further research should be done on the dynamics of guideline development groups, how these interactions are perceived by the group, and their impact on decisions that are made during the course of development (e.g. via participant observation). In addition, research on the implementation of guidelines and the impact of multi-

stakeholder development on successful uptake and use in practice is needed.

Exploring the impacts and possible benefits of stakeholder involvement and co-production would be useful to guideline development organisations globally. A deeper dive into broader stakeholder engagement literature could be done to compare and contrast barriers and facilitators in other health practice and research-related activities as these may apply to specific aspects of guideline development and group work. Little information was found for several stakeholder groups under review, as most studies centred around patients, caregivers, and healthcare providers. It may be that their involvement is more often assumed and therefore less controversial, or they continue to be excluded and understudied. Future studies should explore the engagement of these stakeholder groups, such as policymakers, payers of services, payers of research, peer review editors and product makers. Notably, few studies reported equity characteristics of stakeholders, and future research should consider how best to engage stakeholders from diverse backgrounds (e.g., ethnic or language groups).

**Differences between protocol and review:** None

## Appendices

### Appendix 1. Medline Search Strategy

- 1 Stakeholder Participation/
- 2 patient participation/
- 3 consumer participation/
- 4 Community-Based Participatory Research/
- 5 (coproduction or co-production).ti,ab,kf.
- 6 ((stakeholder\* or advisor\* or reference\* or expert\* or consultation\* or steering) adj2 (group\* or panel\*)).ti,ab,kf.
- 7 ((stakeholder\* or patient\* or consumer\* or public or caregiver\* or care-giver\* or communit\* or citizen\* or user\* or service-user\* or end-user\* or clinician\* or doctor\* or physician\* or nurse\* or policymaker\* or policy-maker\* or funder\* or indust\* or pharmaceutical) adj2 (engag\* or involv\* or input or participat\* or collaborat\*)).ti,ab,kf.
- 8 or/1-7
- 9 practice guidelines as topic/
- 10 Guideline\*.ti.
- 11 Guidance.ti,kf.
- 12 Clinical guideline\*.ti,ab,kf.
- 13 Clinical Practice Guideline\*.ti,ab,kf.
- 14 or/9-13
- 15 8 and 14 (6569)

### Appendix 2. CINAHL Search Strategy

| #   | Query                | Limiters/Expanders            | Last Run Via                                                                                                       |
|-----|----------------------|-------------------------------|--------------------------------------------------------------------------------------------------------------------|
| S11 | S5 AND S10           | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S10 | S6 OR S7 OR S8 OR S9 | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |

|    |                                                                                                                                                                                                                                                                                                          |                               |                                                                                                                    |
|----|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-------------------------------|--------------------------------------------------------------------------------------------------------------------|
| S9 | (clinical OR practice) N1 guideline*                                                                                                                                                                                                                                                                     | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S8 | TI Guidance OR SU Guidance                                                                                                                                                                                                                                                                               | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S7 | TI Guideline*                                                                                                                                                                                                                                                                                            | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S6 | MH Practice Guidelines                                                                                                                                                                                                                                                                                   | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S5 | S1 OR S2 OR S3 OR S4                                                                                                                                                                                                                                                                                     | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S4 | (stakeholder* OR patient* OR consumer* OR public<br>OR caregiver* OR communit* OR citizen* OR user*<br>OR service- user* OR end-user* OR clinician* OR<br>doctor* OR physician* OR nurse* OR policymaker*<br>OR funder* OR industry OR pharmaceutical) N2<br>(engag* OR involv* OR input OR participat*) | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S3 | (stakeholder* OR advisor* OR reference* OR expert*<br>OR consultation OR steering) N2 (group* OR panel)                                                                                                                                                                                                  | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
| S2 | coproduction OR co-production                                                                                                                                                                                                                                                                            | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |

|    |                           |                               |                                                                                                                    |
|----|---------------------------|-------------------------------|--------------------------------------------------------------------------------------------------------------------|
| S1 | MH Consumer Participation | Search modes - Boolean/Phrase | Interface - EBSCOhost<br>Research Databases<br>Search Screen -<br>Advanced Search<br>Database - CINAHL<br>Complete |
|----|---------------------------|-------------------------------|--------------------------------------------------------------------------------------------------------------------|

### Appendix 3. Embase Search Strategy

- 1 Stakeholder Participation/
- 2 patient participation/
- 3 consumer participation/
- 4 Community-Based Participatory Research/
- 5 (coproduction or co-production).ti,ab,kf.
- 6 ((stakeholder\* or advisor\* or reference\* or expert\* or consultation or steering) adj2 (group\* or panel)).ti,ab,kf.
- 7 ((stakeholder\* or patient\* or consumer\* or public or communit\* or citizen\* or user\* or service- user\* or end-user\* or clinician\* or doctor\* or physician\* or nurse\* or policymaker\* or funder\* or industry or pharmaceutical) adj2 (engag\* or involv\* or input or participat\*)).ti,ab,kf.
- 8 or/1-7
- 9 practice guidelines as topic/
- 10 Guideline\*.ti.
- 11 Clinical guideline\*.ti,ab,kf.
- 12 Clinical Practice Guideline\*.ti,ab,kf.
- 13 or/9-12
- 14 8 and 13

### Appendix 4. PsycINFO Search Strategy

- 1 Client Participation/
- 2 Stakeholder/ and Involvement/
- 3 Stakeholder/ and Participation/
- 4 coproduction.ti,ab.
- 5 co-production.ti,ab.
- 6 (stakeholder\* adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 7 (stakeholder\* adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 8 (advisor\* adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 9 (advisor\* adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]



- 10 (reference\* adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 11 (reference\* adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 12 (expert\* adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 13 (expert\* adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 14 (consultation adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 15 (consultation adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 16 (steering adj2 group\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 17 (steering adj2 panel).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 18 (stakeholder\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 19 (stakeholder\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 20 (stakeholder\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 21 (stakeholder\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 22 (patient\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 23 (patient\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 24 (patient\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 25 (patient\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 26 (consumer\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 27 (consumer\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 28 (consumer\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key
- 29 (consumer\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 30 (public adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 31 (public adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 32 (public adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]

- 33 (public adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 34 (caregiver\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 35 (caregiver\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 36 (caregiver\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 37 (caregiver\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 38 (communit\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 39 (communit\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 40 (communit\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 41 (communit\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 42 (citizen\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 43 (citizen\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 44 (citizen\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 45 (citizen\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 46 (user\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 47 (user\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 48 (user\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 49 (user\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key
- 50 (service-user\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 51 (service-user\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 52 (service-user\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 53 (service-user\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 54 (end-user\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 55 (end-user\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]

- 56 (end-user\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 57 (end-user\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 58 (clinician\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 59 (clinician\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 60 (clinician\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 61 (clinician\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 62 (doctor\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 63 (doctor\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 64 (doctor\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 65 (doctor\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 66 (physician\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 67 (physician\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 68 (physician\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 69 (physician\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 70 (nurse\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 71 (nurse\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 72 (nurse\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 73 (nurse\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 74 (policymaker\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 75 (policymaker\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 76 (policymaker\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 77 (policymaker\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 78 (funder\* adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title,

- tests & measures]
- 79 (funder\* adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 80 (funder\* adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 81 (funder\* adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 82 (industry adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 83 (industry adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 84 (industry adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 85 (industry adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 86 (pharmaceutical adj2 engag\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 87 (pharmaceutical adj2 involv\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 88 (pharmaceutical adj2 input).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 89 (pharmaceutical adj2 participat\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 90 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 or 19 or 20 or 21 or 22 or 23 or 24 or 25 or 26 or 27 or 28 or 29 or 30 or 31 or 32 or 33 or 34 or 35 or 36 or 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 or 45 or 46 or 47 or 48 or 49 or 50 or 51 or 52 or 53 or 54 or 55 or 56 or 57 or 58 or 59 or 60 or 61 or 62 or 63 or 64 or 65 or 66 or 67 or 68 or 69 or 70 or 71 or 72 or 73 or 74 or 75 or 76 or 77 or 78 or 79 or 80 or 81 or 82 or 83 or 84 or 85 or 86 or 87 or 88 or 89
- 91 Treatment Guidelines/  
Guideline\*.ti.
- 92 Guidance.ti.
- 93 guidance.id.
- 94
- 95 (clinical adj1 guideline\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 96 (practice adj1 guideline\*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
- 97 91 or 92 or 93 or 94 or 95 or 96
- 98 90 and 97

## Appendix 5. Scopus Search Strategy

(( TITLE-ABS-KEY ( coproduction OR co-production ) ) OR ( TITLE-ABS-KEY ( ( stakeholder\* OR advisor\* OR reference\* OR expert\* OR consultation\* OR steering ) W/2 ( group\* OR panel\* ) ) ) OR ( TITLE-ABS-KEY ( ( stakeholder\* OR patient\* OR consumer\* OR public OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurse\* OR policymaker\* OR policy-maker\* OR funder\* OR industr\* OR pharmaceutical ) W/2 ( engag\* OR involv\* OR input OR participat\* OR collaborat\* ) ) ) ) AND ( ( TITLE ( guideline\* ) ) OR ( TITLE ( guidance ) ) OR ( KEY ( guidance ) ) ) OR ( TITLE-ABS-KEY ( "clinical guideline\*" OR "clinical practice guideline\*" ) ) )

## Appendix 6. Sociological Abstracts Search Strategy

Searched for: (((MAINSUBJECT.EXACT ("Audience participation") OR MAINSUBJECT.EXACT("Participation") OR MAINSUBJECT.EXACT("Stakeholders") OR MAINSUBJECT.EXACT("Community") OR MAINSUBJECT.EXACT("Citizen participation") OR MAINSUBJECT.EXACT("Consumers"))) OR noft((stakeholder\* OR advisor\* OR reference\* OR expert\* OR consultation\* OR steering) NEAR/2 (group\* OR panel\*)) OR noft((stakeholder\* OR patient\* OR consumer\* OR public OR caregiver\* OR care-giver\* OR communit\* OR citizen\* OR user\* OR service-user\* OR end-user\* OR clinician\* OR doctor\* OR physician\* OR nurse\* OR policymaker\* OR policy-maker\* OR funder\* OR industr\* OR pharmaceutical) NEAR/2 (engag\* OR involv\* OR input OR participat\* OR collaborat\*))) AND (MAINSUBJECT.EXACT("Guidelines") OR ti(Guideline\*) OR noft(Guidance\*) OR noft(Clinical NEAR/3 guideline\*))) AND stype.exact("Scholarly Journals")

## ENTREQ Checklist

| No | Item                       | Guide and description                                                                                                                                                                                                                                                                                                                                                                                             | Page no.        |
|----|----------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|-----------------|
| 1  | Aim                        | State the research question the synthesis addresses.                                                                                                                                                                                                                                                                                                                                                              | 154-155         |
| 2  | Synthesis methodology      | Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology ( <i>e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis</i> ).                                                                           | 164-165         |
| 3  | Approach to searching      | Indicate whether the search was pre-planned ( <i>comprehensive search strategies to seek all available studies</i> ) or iterative ( <i>to seek all available concepts until they theoretical saturation is achieved</i> ).                                                                                                                                                                                        | 159, Appendices |
| 4  | Inclusion criteria         | Specify the inclusion/exclusion criteria ( <i>e.g. in terms of population, language, year limits, type of publication, study type</i> ).                                                                                                                                                                                                                                                                          | 155-159         |
| 5  | Data sources               | Describe the information sources used ( <i>e.g. electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists</i> ) and when the searches conducted; provide the rationale for using the data sources. | 159-160         |
| 6  | Electronic Search strategy | Describe the literature search ( <i>e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits</i> ).                                                                                                                                                                   | Appendix 1-6    |
| 7  | Study screening methods    | Describe the process of study screening and sifting ( <i>e.g. title, abstract and full text review, number of independent reviewers who screened studies</i> ).                                                                                                                                                                                                                                                   | 160-161         |
| 8  | Study characteristics      | Present the characteristics of the included studies ( <i>e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions</i> ).                                                                                                                                                                                                                 | 171-            |

| No | Item                    | Guide and description                                                                                                                                                                                                                                                                                                                                                                             | Page no. |
|----|-------------------------|---------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|----------|
| 9  | Study selection results | Identify the number of studies screened and provide reasons for study exclusion ( <i>e.g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications to the research question and/or contribution to theory development</i> ). | 166,167  |
| 10 | Rationale for appraisal | Describe the rationale and approach used to appraise the included studies or selected findings ( <i>e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings</i> ).                                                                                                                                        | 165-166  |
| 11 | Appraisal items         | State the tools, frameworks and criteria used to appraise the studies or selected findings ( <i>e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope [25]; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting</i> ).                                                                                             | 165      |
| 12 | Appraisal process       | Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required.                                                                                                                                                                                                                                                                               | 165      |
| 13 | Appraisal results       | Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale.                                                                                                                                                                                                                                             | 197,198  |
| 14 | Data extraction         | Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? ( <i>e.g. all text under the headings “results /conclusions” were extracted electronically and entered into a computer software</i> ).                                                                                                                                     | 161-164  |
| 15 | Software                | State the computer software used, if any.                                                                                                                                                                                                                                                                                                                                                         | NR       |
| 16 | Number of reviewers     | Identify who was involved in coding and analysis.                                                                                                                                                                                                                                                                                                                                                 | NR       |
| 17 | Coding                  | Describe the process for coding of data ( <i>e.g. line by line coding to search for concepts</i> ).                                                                                                                                                                                                                                                                                               | 164-165  |

| No | Item                 | Guide and description                                                                                                                                                                                                                  | Page no.           |
|----|----------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|--------------------|
| 18 | Study comparison     | Describe how were comparisons made within and across studies ( <i>e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary</i> ).                                            | 164-165            |
| 19 | Derivation of themes | Explain whether the process of deriving the themes or constructs was inductive or deductive.                                                                                                                                           | 164                |
| 20 | Quotations           | Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author's interpretation.                                                           | Table 5-6          |
| 21 | Synthesis output     | Present rich, compelling and useful results that go beyond a summary of the primary studies ( <i>e.g. new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct</i> ). | Table 5-6, 231-257 |



# PROTOCOL: Barriers and facilitators to stakeholder engagement in health guideline development: A qualitative evidence synthesis

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Joanne Khabisa<sup>5</sup> | Pearl Atwere<sup>3</sup> | Elie A. Akl<sup>6</sup> | Pauline Campbell<sup>7</sup> |  
Vivian Welch<sup>8</sup> | Maureen Smith<sup>9</sup> | Reem A. Mustafa<sup>4</sup> | Heather Limburg<sup>10</sup> |  
Leonila F. Dans<sup>11</sup> | Nicole Skoetz<sup>12</sup> | Sean Grant<sup>13</sup> | Tom Concannon<sup>14</sup> |  
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## Abstract

**Background:** There is a need for the development of comprehensive, global, evidence-based guidance for stakeholder engagement in guideline development. Stakeholders are any individual or group who is responsible for or affected by health- and healthcare-related decisions. This includes patients, the public, providers of health care and policymakers for example. As part of the guidance development process, Multi-Stakeholder Engagement (MuSE) Consortium set out to conduct four concurrent systematic reviews to summarise the evidence on: (1) existing guidance for stakeholder engagement in guideline development, (2) barriers and facilitators to stakeholder engagement in guideline development, (3) managing conflicts of interest in stakeholder engagement in guideline development and (4) measuring the impact of stakeholder engagement in guideline development. This protocol addresses the second systematic review in the series.

**Objectives:** The objective of this review is to identify and synthesise the existing evidence on barriers and facilitators to stakeholder engagement in health guideline development. We will address this objective through two research questions: (1) What are the barriers to multi-stakeholder engagement in health guideline development across any of the 18 steps of the GIN-McMaster checklist? (2) What are the facilitators to multi-stakeholder engagement in health guideline development across any of the 18 steps of the GIN-McMaster checklist?

**Search Methods:** A comprehensive search strategy will be developed and peer-reviewed in consultation with a medical librarian. We will search the following databases: MEDLINE, Cumulative Index to Nursing & Allied Health Literature (CINAHL), EMBASE, PsycInfo, Scopus, and Sociological Abstracts. To identify grey

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Hospital Research Institute, Ottawa, Canada

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literature, we will search the websites of agencies who actively engage stakeholder groups such as the AHRQ, Canadian Institutes of Health Research (CIHR) Strategy for Patient-Oriented Research (SPOR), INVOLVE, the National Institute for Health and Care Excellence (NICE) and the PCORI. We will also search the websites of guideline-producing agencies, such as the American Academy of Pediatrics, Australia's National Health Medical Research Council (NHMRC) and the WHO. We will invite members of the team to suggest grey literature sources and we plan to broaden the search by soliciting suggestions via social media, such as Twitter.

**Selection Criteria:** We will include empirical qualitative and mixed-method primary research studies which qualitatively report on the barriers or facilitators to stakeholder engagement in health guideline development. The population of interest is stakeholders in health guideline development. Building on previous work, we have identified 13 types of stakeholders whose input can enhance the relevance and uptake of guidelines: Patients, caregivers and patient advocates; Public; Providers of health care; Payers of health services; Payers of research; Policy makers; Program managers; Product makers; Purchasers; Principal investigators and their research teams; and Peer-review editors/publishers. Eligible studies must describe stakeholder engagement at any of the following steps of the GIN-McMaster Checklist for Guideline Development.

**Data Collection and Analysis:** All identified citations from electronic databases will be imported into Covidence software for screening and selection. Documents identified through our grey literature search will be managed and screened using an Excel spreadsheet. A two-part study selection process will be used for all identified citations: (1) a title and abstract review and (2) full-text review. At each stage, teams of two review authors will independently assess all potential studies in duplicate using a priori inclusion and exclusion criteria. Data will be extracted by two review authors independently and in duplicate according to a standardised data extraction form.

**Main Results:** The results of this review will be used to inform the development of guidance for multi-stakeholder engagement in guideline development and implementation. This guidance will be official GRADE (Grading of Recommendations Assessment, Development and Evaluation) Working Group guidance. The GRADE system is internationally recognised as a standard for guideline development. The findings of this review will assist organisations who develop healthcare, public health and health policy guidelines, such as the World Health Organization, to involve multiple stakeholders in the guideline development process to ensure the development of relevant, high quality and transparent guidelines.

## 1 | BACKGROUND

### 1.1 | The problem, condition or issue

Health guidelines are 'systematically developed evidence-based statements which assist providers, recipients and other stakeholders in making informed decisions about appropriate health interventions'

(WHO, 2003). Guidelines play a crucial role in the delivery of evidence-based medicine. The systematic examination of evidence promoted by evidence-based medicine is done through a comprehensive search of the literature, critical appraisal of the quality of the evidence and interpretation of the findings in light of patients' preferences and societal values (Guyatt, 2008). These guidelines assist health practitioners, patients, caregivers, policymakers and



other stakeholders to make informed decisions about health practice, public health and health policy. It is increasingly recognised that the engagement of multiple stakeholders in guideline development improves recommendation relevancy, uptake, implementation and sustainability (Esmail, 2015; Gagliardi, 2012; Moulding, 1999).

Stakeholders are 'any individual or group who is responsible for or affected by health- and healthcare-related decisions that can be informed by research evidence' (Concannon, 2012). This broad definition includes, but is not limited to, patients, caregivers, the public, patient advocates, providers (e.g., nurses), policymakers (e.g., health ministries), payers of health services (e.g., insurers, public payers) and health research (e.g., national research institutes), purchasers (e.g., employers), researchers, product makers (e.g., drug makers) and peer review editors. Several entities, including the World Health Organization (WHO), the Canadian Institute for Health Research (CIHR), the National Institute for Health and Care Excellence (NICE) and the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) Working Group recommend the inclusion of stakeholder groups in the guideline development and implementation process (AKI, 2017; CIHR, 2014; NICE, 2014; WHO, 2003). The Guidelines International Network (GIN) McMaster Checklist for Guideline Development, the global standard for guideline development, identifies 'consumer and stakeholder involvement' as Step 6 in the guideline development process (Schünemann, 2014).

There is a growing body of research into the methods for engaging stakeholders in guideline development (Armstrong, 2017; Cluzeau, 2012), as well as approaches to measuring the impact of stakeholder engagement in guideline development (Cottrell, 2014; Ray, 2017). For example, a 2017 review of the incorporation of patients' values and preferences in guidelines included guidance documents from 56 different organisations (Selva et al., 2017). However, there remains a need for the development of comprehensive, global, evidence-based guidance for stakeholder engagement in health guideline development that brings together the vast literature amassed to date. The Multi-Stakeholder Engagement Consortium (MuSE Consortium) was established in 2015 to advance methods and approaches used in stakeholder engaged health research. The consortium addresses research of all kinds—including reviews and guidelines – and includes more than 80 researchers, trainees and stakeholders from Australia, Brazil, Canada, Germany, Italy, Lebanon, the Netherlands, the Philippines, Switzerland, the UK and the USA. The team membership of MuSE includes researchers, policymakers, guideline developers, research funders, clinicians, patients, patient representatives and policymakers from various organisations including the Agency for Healthcare Research and Quality (AHRQ), the Campbell Collaboration, Cochrane, GRADE Working Group, Public Health Agency of Canada, Patient-Centered Outcomes Research Institute (PCORI), Research and Development (RAND) Corporation, the World Health Organization (WHO), Joanna Briggs Institute (JBI) and multiple universities (Petkovic, 2020). As part of the consortium's work, a working group was formed to develop global guidance on stakeholder engagement in guideline development according to the

18-step GIN-McMaster Checklist. The working group set out to conduct four concurrent systematic reviews to summarise the evidence on: (1) existing guidance for stakeholder engagement in guideline development, (2) barriers and facilitators to stakeholder engagement in guideline development, (3) managing conflicts of interest in stakeholder engagement in guideline development and (4) measuring the impact of stakeholder engagement in guideline development. This protocol addresses the second systematic review in the series.

## 1.2 | Definitions

For the purpose of this review, we define:

- *Guidelines* as 'systematically developed evidence-based statements which assist providers, recipients and other stakeholders in making informed decisions about appropriate health interventions' (WHO, 2003).
- *Stakeholders* as 'any individual or group who is responsible for or affected by health- and healthcare-related decisions that may be informed by research evidence' (Concannon et al., 2012).
- *Engagement* as an approach to ensure the contribution of stakeholders towards the development of the guideline, completion of any of the stages of the guideline, or dissemination of the guideline and its recommendations (Frank, 2020; Pollock, 2018).
- *Levels of engagement* as the intensity of involvement in the guideline development process (Oliver, 2008; Pollock, 2019). We operationalise two levels for the purpose of this review: (1) advisory/feedback or (2) participation in decision-making or knowledge translation.

## 1.3 | Description of the phenomena of interest

Stakeholder engagement in guideline development is the approach to gather input or contribution from stakeholders 'towards the development of a guideline, completion of any stages of a guideline, or dissemination, uptake or evaluation of a guideline and its recommendations' (Pollock, 2018). The term 'stakeholders' is inclusive of multiple individuals within a single stakeholder group (such as several patients) and multiple stakeholders across several stakeholder groups (such as patients, policymakers and providers of care). Engagement refers to a meaningful and active partnership among stakeholder groups (CIHR, 2019; Staniszewska, 2017) to develop, complete or disseminate a guideline. Other terms for engagement include 'public involvement', 'consumer engagement', 'Patient and Public Involvement (PPI)', 'coproduction', 'co-creation', 'activation' and 'involvement'.

Stakeholder engagement in research can occur at different levels of intensity, ranging from low-intensity communication (i.e., stakeholders are informed of research processes) up to high-intensity

coproduction (i.e., stakeholders are equal members of the research team and participate in shared decision-making) (Concannon, 2012). From previous work (Crowe, 2017; Oliver, 2008; Pollock, 2019), we identify two levels of engagement in guideline development, namely (1) advisory/feedback, and (2) participation in decision-making or knowledge translation. These levels were selected for our operational use, but we acknowledge that within these, there may be different levels.

There are many activities required for effective stakeholder engagement in guideline development (see Figure 1). The engagement of stakeholders from multiple disciplines or specialties is typically recommended to take account of a wide range of opinions. Indeed, the greater diversity in stakeholders' background increases the knowledge and skills available to make decisions (Oliver, 2018). However, the ability to gather and apply input from multiple stakeholders in a way that meaningfully reflects and addresses stakeholders' interests is not easily achieved, as increased diversity may also reduce group cohesiveness (Oliver, 2018). In their review of theoretical literature, Brodbeck and colleagues (Brodbeck, 2007) proposed that groups can outperform individual decision makers under conditions which promote sharing of knowledge and mutual learning. Oliver and colleagues (Oliver, 2018) built on this model, and suggested that these conditions include diverse group membership, skilled facilitation, time for knowledge sharing, following formal consensus processes, and integrating divergent perspectives. Stakeholder engagement is a complex process, and achieving these conditions requires both individual and organisational adoption. Organisational cultures and relationships act as a backdrop to these stakeholder engagement activities (Oliver, 2018). There are a

multitude of individual- and organisational-level factors that can affect a stakeholder's ability to engage in the guideline development process, or a guideline developer's ability to effectively engage them. Barriers to stakeholder engagement are defined as any variable, condition or factor that may impede stakeholder engagement in guideline development, while facilitators are any variable, condition or factor that may promote stakeholder engagement in guideline development. Such determinants may include knowledge, skills, time and resources, for example, and may occur across one or more steps of guideline development.

## 1.4 | Why it is important to do this review

With increased recognition of the value of stakeholder engagement in guideline development, there is a need for expert opinion and evidence-based guidance on when and how to engage stakeholders. This review of barriers and facilitators to stakeholder engagement in guideline development will help fill this gap. It will build on existing reviews of barriers and facilitators, synthesise the current evidence-base, and examine a broad range of stakeholder groups.

To date, several systematic reviews have been conducted on the topic of stakeholder engagement in guideline development, but few are explicitly focused on barriers and facilitators to engagement. Oxman and colleagues (Oxman et al., 2006) conducted a series of reviews to advise WHO to ensure that health care recommendations are informed by the best available research evidence. Relevant reviews in this series included group composition (Fretheim, 2006a), group processes (Fretheim, 2006b) and consumer involvement

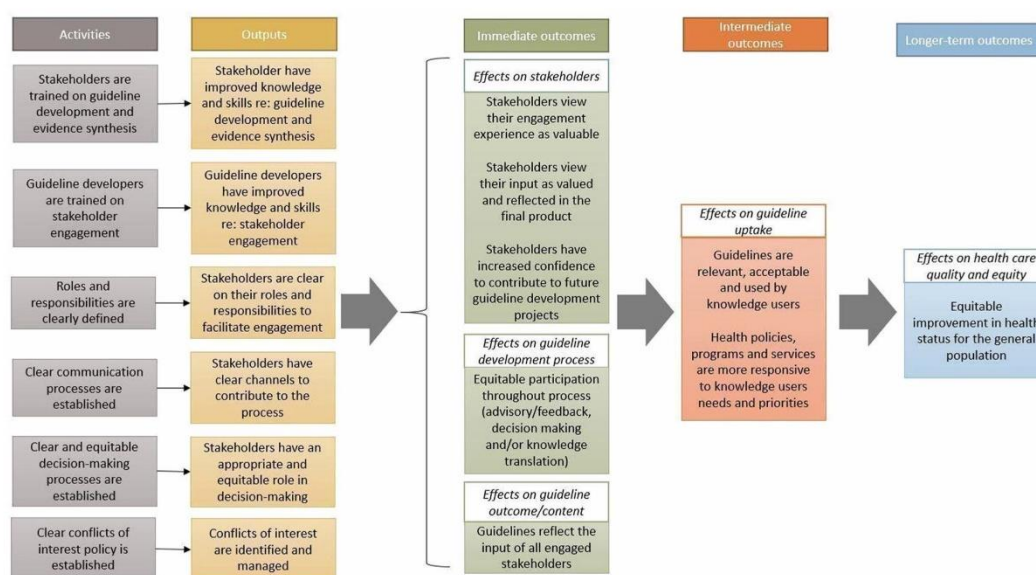


FIGURE 1 Logic model of the effects of stakeholder engagement in guideline development



(Schünemann, 2006). However, these reviews were not systematic nor exhaustive, and prioritised existing syntheses over primary empirical studies. Relatedly, Oliver and colleagues (Oliver, 2018) synthesised theoretical literature to learn about the structure, processes and environment of committees generally, which has applications for health guideline development.

Much of the existing barrier and facilitator syntheses focus specifically on patients. Légaré and colleagues (Légaré, 2011) reviewed published and unpublished literature regarding the effectiveness of patient and public engagement programs in clinical practice guidelines and described the barriers and facilitators to these programs. Barriers included recruiting challenges, lack of representation of certain patient and public groups, and participants' lack of familiarity with medical and scientific terminology. Facilitators included the provision of training and support. Similarly, Grant and colleagues (Grant, 2018) conducted a rapid review and qualitative evidence synthesis to make inferences about how feasible online methods for patient engagement would be during clinical practice guideline development. The authors highlighted important considerations for patient and caregiver participation related to skills, time and resources. Notably, both of these reviews only considered patient and public involvement and did not consider the engagement of other stakeholder groups such as healthcare providers or policymakers, for example. Recently, Selva and colleagues (Selva, 2017) reviewed guidance documents for developing clinical guidelines to assess how these documents address the incorporation of patients' views. They identified the need for additional guidance in this area.

Cluzeau and colleagues (Cluzeau, 2012; Kelson, 2012; Kunz, 2012) conducted a literature review and workshop to answer six questions related to stakeholder engagement in guideline development, including identification of the potential barriers and facilitators to integrating stakeholder involvement. They defined stakeholders as 'all those who have a legitimate interest in a guideline. They include healthcare professionals, patients and caregivers, public and private funding bodies, managers, employers and manufacturers'. They noted that integrating different, sometimes competing, stakeholder perspectives can be challenging. Other barriers included issues with potential bias among stakeholder views, and the costly nature of the process. In contrast, they found that educating stakeholders and ensuring effective communication enabled effective stakeholder engagement.

In sum, there is a lack of systematic reviews which have synthesised barriers and facilitators to multi-stakeholder engagement in health guideline development. The results of this review will be used to inform the development of guidance for multi-stakeholder engagement in guideline development and implementation. This guidance will be official GRADE (Grading of Recommendations Assessment, Development and Evaluation) Working Group guidance. The GRADE system is internationally recognised as a standard for guideline development (Guyatt, 2008). The findings of this review will assist organisations who develop healthcare, public health and health policy guidelines, such as the World Health Organization, to involve multiple stakeholders in the guideline development process to ensure the development of relevant, high quality and transparent guidelines.

## 2 | OBJECTIVES

The objective of this review is to identify and synthesise the existing evidence on barriers and facilitators to stakeholder engagement in health guideline development. We will address this objective through two research questions:

1. What are the barriers to multi-stakeholder engagement in health guideline development across any of the 18 steps of the GIN-McMaster checklist?
2. What are the facilitators to multi-stakeholder engagement in health guideline development across any of the 18 steps of the GIN-McMaster checklist?

## 3 | METHODS

The methods for this systematic review will follow the *Cochrane Handbook for Systematic Reviews of Interventions* and the *Handbook for Synthesizing Qualitative Research*, as appropriate (Higgins, 2019; Sandelowski, 2007). These methods were developed by a multi-stakeholder research team that engaged investigators, trainees, providers of care, policymakers and patient representatives.

### 3.1 | Criteria for including and excluding studies

#### 3.1.1 | Types of study designs

We will include empirical qualitative and mixed-method primary research studies which qualitatively report on the barriers or facilitators to stakeholder engagement in health guideline development. This includes qualitative research studies using common methods such as interviews, focus groups, or surveys to collect participant experiences; case studies of existing programmes; and process evaluation studies. We will also include published guidelines which report lessons learned only if these statements are derived from empirical qualitative methods and the details of these methods are available in a published report. Mixed-method and intervention studies (randomised or non-randomised designs) will be eligible only if they apply qualitative methods and qualitatively report barriers and facilitators to engagement, independent of their quantitative findings.

Conference abstracts, commentaries, editorials, protocols, systematic reviews, meta-ethnographies and other literature reviews will be excluded.

#### 3.1.2 | Population of interest

The population of interest is stakeholders in health guideline development. We define stakeholders as 'any individual or group who is responsible for or affected by health- and healthcare-related decisions' (Concannon, 2012). Building on previous work, we have

identified 13 types of stakeholders whose input can enhance the relevance and uptake of guidelines (Concannon, 2012, 2019; Tugwell, 2006). We will include studies which report on the barriers or facilitators to engagement of one or several of these stakeholder groups at any level of intensity at any step of the guideline development process. For comparisons with previously published stakeholder frameworks, please see Supporting Information Appendix 7. For the purposes of this review, have grouped them as follows:

- Patients, caregivers and patient advocates
- Public
- Providers of health care
- Payers of health services
- Payers of research
- Policy makers
- Program managers
- Product makers
- Purchasers
- Principal investigators and their research teams, and
- Peer-review editors/publishers

We recognise that within each of these categories, there is diversity and heterogeneity in types of stakeholders. For example, we recognise that patients and caregivers have differing characteristics and experiences, and their engagement brings diverse value to guideline development. The categories outlined above are for operational purposes and are based on prior research (Concannon, 2012, 2019; Tugwell, 2006). Studies which consider a single group of stakeholders or multiple groups of stakeholders will be eligible for inclusion. If a study includes multiple stakeholder groups, findings will be coded for each stakeholder group separately.

### 3.1.3 | Phenomena of interest

Eligible studies must describe stakeholder engagement at any of the following steps of the GIN-McMaster Checklist for Guideline Development (Supporting Information Appendix 9) (Schünemann, 2014):

1. Organization, budget, planning and training
2. Priority-setting
3. Guideline group membership
4. Establishing guideline group processes
5. Identifying target audience and topic selection
6. Consumer and stakeholder involvement
7. Conflict of interest considerations
8. Question generation
9. Considering importance of outcomes and interventions, values, preferences and utilities
10. Deciding what evidence to include and searching for evidence
11. Summarising evidence and considering additional information
12. Judging quality, strength or certainty of body of evidence

13. Developing recommendations and determining their strength
14. Wording of recommendations and of considerations about implementation, feasibility and equity
15. Reporting and peer review
16. Dissemination and implementation
17. Evaluation and use
18. Updating

This checklist was selected because it represents the global standard for guideline development. Eligible studies must report primary data in their results on barriers and facilitators to stakeholder engagement at at least one step of the checklist. Studies which report on barriers and facilitators at one step, multiple steps, or across all steps will be considered for inclusion.

### 3.1.4 | Outcomes of interest

The primary outcomes of interest are any barriers and facilitators to stakeholder engagement in health guideline development.

Barriers are defined as *any variable or condition that impedes stakeholder engagement in guideline development or implementation*.

Facilitators are defined as *any variable or condition that promotes stakeholder engagement in guideline development or implementation*.

For qualitative evidence, we will analyse first- and second-order constructs related to stakeholders' perceptions and experiences of barriers and facilitators to their engagement in guideline development and implementation. These include participant quotations from interviews or focus groups, excerpts or quotations from documentary analysis, narrative descriptive summaries, author hypotheses, explanations and recommendations, themes and sub-themes. First order-constructs are defined as participant quotes and participant observations, while recognising that in secondary analysis these represent the participants' views as selected by the study authors in evidencing their second-order constructs. Second-order constructs are study authors' themes/concepts and interpretations.

### 3.1.5 | Setting

We will place no restrictions on geographic setting. Studies focused on health guideline development from any country in the world are eligible for inclusion.

### 3.1.6 | Language

No language restrictions will be applied.

### 3.1.7 | Publication date

No publication date restrictions will be applied.

### 3.2 | Search methods for identification of studies

This review is part of a series of four reviews conducted by the MuSE working group on stakeholder engagement in guideline development. As such, one comprehensive search strategy was developed and peer-reviewed in consultation with a medical librarian in accordance with PRESS guidelines (McGowan, 2016). A second medical librarian reviewed the search strategy.

#### 3.2.1 | Electronic databases

We will search the following databases: MEDLINE (OVID) (Supporting Information Appendix 1), Cumulative Index to Nursing & Allied Health Literature (CINAHL; EBSCO) (Supporting Information Appendix 2), EMBASE (OVID) (Supporting Information Appendix 3), PsycInfo (OVID) (Supporting Information Appendix 4), Scopus (Supporting Information Appendix 5) and Sociological Abstracts (Supporting Information Appendix 6). Limits will not be placed on date, study design or language.

#### 3.2.2 | Searching other resources

To identify grey literature, we will search the websites of agencies who actively engage stakeholder groups such as the AHRQ, Canadian Institutes of Health Research (CIHR) Strategy for Patient-Oriented Research (SPOR), INVOLVE, the National Institute for Health and Care Excellence (NICE) and the PCORI. We will also search the websites of guideline-producing agencies, such as the American Academy of Pediatrics, Australia's National Health Medical Research Council (NHMRC) and the WHO. We will invite members of the team to suggest grey literature sources and we plan to broaden the search by soliciting suggestions via social media, such as Twitter.

#### 3.2.3 | Citations and reference lists

Backward and forward citation tracking will be performed on included articles to identify further eligible studies. We will review reference lists of relevant reviews to identify eligible primary studies for inclusion.

#### 3.2.4 | Contacting experts

We will contact authors of included studies to ask for suggested studies. We will also ask our MuSE Consortium members for potentially eligible primary studies that were not identified through the database search.

#### 3.2.5 | Screening of studies

All identified citations from electronic databases will be imported into Covidence software for screening and selection. Documents

identified through our grey literature search will be managed and screened using an Excel spreadsheet. A two-part study selection process will be used for all identified citations: (1) a title and abstract review and (2) full-text review. At each stage, teams of two review authors will independently assess all potential studies in duplicate using a priori inclusion and exclusion criteria. We will resolve any disagreements through discussion or, if required, we will consult a third review author. We will produce a PRISMA flow diagram which reports the number of studies included and excluded at each stage. Reasons for exclusion will be provided for all studies assessed at full-text.

### 3.3 | Description of methods used in primary research

Relevant studies will have employed a qualitative or mixed-method design. For example:

- Brouwers et al. (2017) (mixed-methods; Brouwers, 2017). Cancer patients, survivors, family members and caregivers were recruited to explore optimal approaches to patient and caregiver engagement in the development of cancer practice guidelines. Participants attended a workshop, completed a survey, or participated in a telephone interview. Quantitative data were analysed using Microsoft Excel, and qualitative data were analysed using thematic analysis. Authors report on barriers and facilitators to engaging in guideline development.
- Atkins et al. (2013) (qualitative; Atkins, 2013). Members of three National Institute of Health and Clinical Excellence (NICE) advisory groups on acute physical, mental and public health were interviewed to investigate how they conceptualise evidence and experience the process. Participants were interviewed at the beginning and end of the life of the group. Interview transcripts were analysed using thematic and content analysis to identify main themes.

### 3.4 | Criteria for determination of independent findings

#### 3.4.1 | Multiple reports of the same study

If a guideline development process is evaluated by more than one publication and is published in multiple reports, we will only include the most recent report or any additional report with unique outcome assessments (e.g., secondary analysis reports). If several reports of the same guideline process are identified and all publications report on different outcomes, each individual report must meet eligibility criteria for inclusion. Eligible reports of the same study will be analysed as a single study. Information on study sample sizes, guideline details, grant numbers and so on will be used to identify multiple reports from single studies and multiple studies in single



reports. The authors of the reports will be contacted if it is unclear whether reports and studies provide independent findings.

### 3.4.2 | Multiple studies in single reports

If more than one study is described in a single report, each study within the report will be coded separately.

### 3.4.3 | Multiple studies involving the same guideline development team

Reports published by the same guideline development team are likely to follow similar stakeholder engagement practices, and so multiple reports published by a single guideline team will be aggregated.

## 3.5 | Details of study coding categories

Data will be extracted by two review authors independently and in duplicate according to a standardised data extraction form (see Supporting Information Appendix 8). Disagreements will be discussed and resolved by a third reviewer. The data extraction form will be pilot tested by two review authors and coding categories will be clarified iteratively, as needed. Data will be extracted on basic study characteristics and methods including:

- Study objective
- Study setting
- Type of guideline
- Population and participant characteristics, including categorisation according to stakeholder group and equity-relevant characteristics according to PROGRESS-Plus (O'Neill, 2014).
- Recruitment methods
- Data collection methods
- Analysis methods

In addition, we will extract information on study:

- Limitations
- Recommendations
- Conclusions
- Funding
- Conflict of Interest

For qualitative outcomes, we will extract data using codes adopted from the Theoretical Domains Framework (Atkins, 2017). We will extract stakeholders' perceptions and experiences regarding the barriers and facilitators to their engagement in health guideline development from the results and discussion sections, including participant quotations from interviews or focus groups, excerpts or quotations from documentary analysis, narrative descriptive

summaries, author hypotheses, explanations and recommendations, themes and sub-themes.

Discrepancies in the data extraction process will be resolved by a third reviewer.

### 3.5.1 | Dealing with missing data

Missing data is an inherent component of the qualitative research process. Since meanings and interpretations are the outputs of qualitative studies, data are always missing because there are some questions that respondents do not want to answer, others they circumvent, and aspects of their experiences which may not be reported (Singh, 2013). In such situations, the silence on these issues in itself becomes powerful data. Our findings will be mapped against the GIN-McMaster Checklist and we will report which steps of the checklist do have supporting qualitative evidence and those which do not, thereby highlighting gaps in evidence and opportunities for new qualitative inquiry.

## 3.6 | Critical appraisal

Two review authors will assess the methodological rigour of all included studies independently and in duplicate. We will assess the quality of qualitative studies and the qualitative components of mixed method studies using the Critical Appraisal Skills Programme (CASP) qualitative appraisal research tool (Singh, 2013). Discrepancies in the critical appraisal process will be resolved by a third reviewer.

## 3.7 | Treatment of qualitative research

### 3.7.1 | Identification of key findings

Our included studies will employ a qualitative approach to inquiry. We will use the 'best fit' framework method as a systematic and flexible approach to analysing the qualitative data (Booth, 2015; Carroll, 2013; Gale, 2013). Framework-based synthesis, using the 'best fit' method, is a highly pragmatic and useful strategy for analysing a range of questions (Dixon-Woods, 2011) and is supported by guideline development literature (Flemming, 2019). Framework analysis is a five-stage process that includes familiarisation with the data, identifying a thematic framework, indexing (applying the framework), charting and mapping and interpretation (Spencer & Ritchie, 1994).

Implementing new or changing existing stakeholder engagement practices, in organisations who develop guidelines, requires a change in individual and collective behaviour. Changing behaviour requires an understanding of the influences on behaviour in the context in which they occur. We selected the Theoretical Domains Framework (TDF) for extracting and analysing our barriers and facilitator data (Atkins, 2017). Developed through a multidisciplinary consensus



approach and subsequent validation, TDF consolidates overlapping behavioural theories into 14 domains encompassing 84 theoretical constructs, and provides a theoretical lens through which to view the cognitive, affective, social and environmental influences on behaviour and implementation.

Data will be analysed deductively, using the TDF domains to generate a framework of preliminary themes. Whilst the domains are purposively designed to be broad groupings of the possible factors to influence behaviour, the intent is to explore the important domains in further detail. Data will be coded independently by two reviewers. Disagreements will be resolved by discussion. The entire review team will be involved in developing key findings.

### 3.7.2 | Mapping of key findings

All identified key findings will be mapped to the 18 topics (146 steps) of the guideline development process outlined by the GIN-McMaster checklist (Shunemann 2014). We will present this information as a matrix indicating the barriers and facilitators that exists for each stakeholder group and for each step of guideline development. The findings of this mapping exercise will be combined with the results of the other reviews in this series (Khabisa, 2022; Petkovic, 2022) to develop international guidance on stakeholder engagement in guideline development.

## 3.8 | Certainty of evidence

We will use the Confidence in the Evidence from Reviews of Qualitative research (CERQual) tool to assess the confidence of our findings. This tool is a method for assessing the strength of qualitative review evidence, similar to how the GRADE approach assesses the strength of quantitative evidence (Lewin et al., 2018). CERQual bases the evaluation on four criteria: (a) methodological limitations of included studies supporting a review finding, (b) the relevance of included studies to the review question, (c) the coherence of the review finding and (d) the adequacy of the data contributing to a review finding. Final CERQual assessments will be presented as a CERQual Evidence Profile.

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## CONTRIBUTIONS OF AUTHORS

Conceiving the review: PT, VW, TC, JP  
Designing the review: JP, AR, VW, PT, TC  
Coordinating the review: AR, OM, JP

Writing the protocol: AR, OM, JP, PA, JK, LL

Providing general advice on the review: EAA, MS, RM, VW, LD, NS, PC, SG

Securing funding for the review: PT, VW, JP

## DECLARATIONS OF INTEREST

None.

## PUBLISHED NOTES

None.

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## SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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## CHAPTER 5: ARTICLE 3. Principles and strategies for interest-holder engagement in health guideline development

### Chapter Overview

This Chapter presents a qualitative study that explores the principles and strategies that interest-holders believe are important to promote their engagement in the guideline development process. This article has been prepared for publication in the *BMC Medical Research Methodology* collection “Inclusive methodological awareness for equity and diversity” (submission deadline August 30, 2024)

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## **5.1 ABSTRACT**

**Background:** Guidelines are statements or recommendations that help interest-holders make decisions about clinical care or health policy. Engaging a wide range of interest-holders, such as patients, providers, policymakers and members of the public in the guideline development process can help ensure that guidelines are fit-for-purpose. The MuSE Consortium is developing

guidance for the engagement of interest-holders throughout the guideline development process in the form of an extension of the Guidelines International Network (GIN)-McMaster Guideline Development Checklist (GDC). The objective of this study is to inform this guidance by identifying principles and strategies to promote interest-holder engagement.

**Methods:** We conducted interviews with 43 individuals from 10 different interest-holder groups and 15 different countries. We used a framework analysis to thematically analyze findings and identify principles and strategies relevant for engaged guideline development.

**Results:** Guideline development initiatives may better engage interest-holders by shifting from vertical to horizontal power structures, emphasizing collaboration and shared decision-making. We identified a need for epistemic justice, promoting fairness and equality in knowledge production and validation, particularly for patients and members of the public. Representation is a crucial key issue, necessitating diverse perspectives in guideline development groups. Strategies include involving interest-holders in governance, setting clear expectations and timelines, fair compensation, and meaningful engagement throughout the process. Dissemination activities should extend beyond academic publications, empowering all interest-holders to contribute to activities. While engagement is desirable, limitations may arise in emergency contexts or resource-constrained settings.

**Conclusions:** Guideline developers may need to make pragmatic decisions as to who they engage in guideline development and how. Capacity strengthening in low- and middle-income countries may help address current disparities in engagement in guideline development. Future research should explore issues around representativeness of interest-holders.

**Keywords:** Stakeholder engagement, interest-holders, Patient and public involvement, guideline development

## 5.2 INTRODUCTION

Guidelines are systematically developed statements to assist decision-makers such as healthcare providers, patients and policymakers make decisions about clinical care, public health and health systems (IOM, 2011). These statements, termed ‘recommendations’, are typically developed through rigorous evidence synthesis and assessments of the benefits and harms of health interventions (IOM, 2011). Since these recommendations about health and treatment services eventually affect people’s lives, engagement in health guideline and policy development is considered a component of people’s right to health (OHCHR, 2008; Vahdat, et al., 2014). Indeed, guideline development organizations such as the World Health Organization (WHO), National Institute for Health and Care Excellence (NICE) and the Guidelines International Network (GIN) have recognized the important role of engaging a range of interest-holders in the guideline development process (WHO, 2014, NICE, 2014, GIN, 2021). “Interest-holders” (previously known as “stakeholders”; Akl et al., in progress) are individuals and groups with legitimate interests that should be considered in the context of health research. These interests draw their legitimacy from the fact that these people are responsible for or affected by health- and healthcare-related decisions that can be informed by research evidence (Akl, in progress; Concannon et al., 2012). The MuSE Consortium, an international group of over 160 individuals interested in engagement in research and guidelines, has identified 10 types of interest-holders that are relevant to the context of health guideline development. These include patients and caregivers, payers of health research, purchasers of health services, peer reviewed journal editors, policymakers, principal investigators, product makers, program managers, providers of care, and members of the public (Petkovic et al., 2023).

A rigorous guideline development process follows a set of principles including multidisciplinary collaboration and a standardized approach for synthesizing evidence and moving from evidence to recommendations (Garbi, 2021, De Leo et al., 2023). There are manuals and checklists for guideline development such as the Guidelines International Network (GIN)-McMaster Guideline Development Checklist (GDC) (Schunemann et al., 2014), which is a comprehensive checklist of items that guideline developers could consider for all stages of the guideline enterprise, from the planning and formulation of recommendations, to their



implementation, evaluation and updating. However, there is a lack of guidance on which interest-holders should be engaged in guideline development, when during the process they should be engaged, and what principles of engagement should be followed for this engagement to be meaningful. The MuSE Consortium aims to address this gap by developing guidance for engagement of interest-holders throughout the guideline development process in the form of an extension for engagement of the GIN-McMaster GDC (Petkovic et al., 2020).

Recent work completed by the MuSE Consortium identified gaps in published evidence on the best ways to engage with interest-holders in health guideline development (Petkovi et al., 2022; Magwood et al., 2022). Most of the identified evidence was focused on patient and public engagement, with very little literature available for other groups such as policymakers, program managers, purchasers, peer review journal editors, product makers, and payers of research. Additionally, there was a clear lack of evidence related to the engagement of people who experience health inequities and are from low-income settings (Magwood et al., 2022). Similarly, a review by Pinho-Gomes and colleagues (2022) summarized the available evidence on principles, values, frameworks, and strategies underpinning patient and public engagement in guideline development, but not broader interest-holder engagement. Furthermore, while they did identify some broad themes such as representation, transparency, relevance, equity, fairness, and reconciling different types of knowledge, the frameworks often failed to explicitly link these principles with the guideline development stages through actionable engagement strategies.

To complement the guidance from the MuSE Consortium on engagement in health guideline development and to promote its operationalization, it is critical to identify principles and strategies for engaging interest-holders in guideline development that are acceptable to them. This article addresses this need by aiming to identify principles and strategies to promote engagement in the development of health guidelines from the perspective of a broad range of interest-holders.

## 5.3 METHODS

To inform guidance for the engagement of interest-holders in health guideline development, the MuSE Consortium conducted a series of reviews, an online survey, key informant interviews and a consensus process. The methods of the project have been reported in the project protocol (Petkovic et al., 2020) and extension paper (Petkovic et al., submitted #2). This paper reports on findings from the key informant interviews addressing the following research question: What are interest-holders' perspectives regarding principles and strategies to promote their engagement in the development of health guidelines?

### **Research team and reflexivity**

The MuSE Consortium is coordinated by a core team who manage the daily tasks of the group, secure and manage funding for project activities, and communicate frequently with all 160+ members of the MuSE Consortium. The core team worked with members of the MuSE Consortium to recruit 2–4 individuals from each of the above-mentioned 10 groups of interest-holders to co-lead the development of the guidance. Co-leads were selected for their expertise relevant to their interest-holder group (for example, lived experience of a health condition or research experience) and experience with health guidelines. We aimed to ensure co-leads were balanced between high- and low- and middle-income countries. In total, 26 co-leads participated in decision-making throughout critical stages of the guidance development (Petkovic et al., submitted). The specific role of co-leads in this study is described further under the section 'data collection'.

The coordination of interviews was led by O.M., a female doctoral candidate with previous experience with qualitative research projects and health guideline development. She worked with the co-leads to develop interview guides, obtained institutional ethics approval, recruited participants, collected data through interviews, and led the analysis. O.M. had previously met some (n=8) interview participants in a professional capacity (e.g., conference, workshop, guideline panel) but did not have any established relationships with them. Interviews were also conducted by a second female doctoral candidate (L.L.), a female research associate (J.K.) and a male medical student (A.L.). All additional interviewers were provided with

orientation to the interview guides and consent forms prior to conducting interviews. This included reasons for doing the research and how the interview data would be used. None of the additional interviewers had relationships with the participants they interviewed.

## Study setting and recruitment

All components of this pragmatic research study occurred virtually, allowing for the recruitment of participants from across the globe. We recruited individuals who self-identified as belonging to any of the 10 groups of interest-holders listed in Table 1. Participants were recruited through recommendations made by the MuSE Consortium network, snowball sampling, and social media. Recruitment occurred between May 2022 and November 2023. We prioritized recruitment of groups who co-leads identified as playing the biggest roles in guideline development (e.g., patients, members of the public, providers of care, principal investigators).

**Table 1. Interest-holder groups to engage in guideline development (Petkovic et al., 2023)**

| Interest-holder group                                           | Description                                                                                                                                                                         |
|-----------------------------------------------------------------|-------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| Patients, patient caregivers, patient advocates/organizations   | Those with lived experience with the condition of interest or who care for or advocate on behalf of those with lived experience                                                     |
| Payers of health research                                       | Individuals and organizations that fund research projects, such as government funders, industry funders, foundations                                                                |
| Payers / Purchasers of Health Services                          | Individuals, organizations and entities that pay for health services                                                                                                                |
| Peer Reviewed Journal Editors                                   | Those who set journal policy on guidelines and manage the peer review process and editing                                                                                           |
| Policymakers                                                    | Individuals, organizations and entities that craft public or private policy (on health) at any level of government                                                                  |
| Principal Investigators and all members of the of research team | Individuals, organizations, and associations that conduct or advocate health research                                                                                               |
| Product makers                                                  | Individuals working for companies that manufacture pharmaceuticals, medical devices, medical procedures, health technologies, or for profit educational and behavioral packages     |
| Program managers                                                | Managers/directors who plan, lead, oversee, or deliver any program that provides public health, community services, or clinical care (e.g. budgeting, hiring, staffing, organizing, |

|           |                                                                                                                                                                              |
|-----------|------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
|           | coordinating, reporting)                                                                                                                                                     |
| Providers | Persons and their professional associations who provide health care in a professional capacity and allowed by regulatory bodies to provide a health care service             |
| Public    | Individuals in the general population of a defined geographic area excluding patients, caregivers, and health professionals living or working with the condition of interest |

## Data collection

We co-developed interview guides with the co-leads from all MuSE interest-holder groups (Appendix 1 presents an example interview guide, designed for interviews with patients, patient caregivers, and patient advocates/organizations). All co-leads had the opportunity to develop questions specific to the key issues for their own interest-holder group. The interview guides were developed iteratively over a series of meetings and email exchanges. Each interview guide included introductory questions to learn about the participant's role and experience with guideline development. Additionally, participants were asked about the GIN-McMaster Guideline Development Checklist (Schunemann et al., 2014). The checklist comprises 18 overarching topics. For each topic, respondents indicated whether or not they wanted to be engaged as decision-makers or as advisors providing feedback, the results of which are described elsewhere (Petkovic, submitted; Magwood in progress). Participants were also asked about conceptual principles and actionable strategies for working with them to develop health guidelines. Four team members (OM, LL, JK, AYSL) conducted interviews virtually using Zoom for a duration of 30-75 minutes. With participants' consent, the interviews were recorded and transcribed verbatim using an automated transcription app (Otter.ai). Transcripts were then imported into NVivo (version 14, Lumivero) for analysis. We anonymized all transcripts by assigning a unique identifier to each interviewee.

## Data analysis

We applied the principles of framework analysis to analyze the qualitative data from interviews. Framework analysis is a five stage process of familiarization with the data, identifying

a thematic framework, indexing (applying the framework), charting and mapping, and interpretation (Ritchie & Spencer, 1994). The framework method is not aligned with a particular epistemological, philosophical, or theoretical approach, making it appealing for pragmatic research (Gale et al., 2013). It can be adapted for use with deductive, inductive, or combined types of qualitative analysis. To guide the analysis, we selected the subcategories of principles and strategies reported by Hoekstra and colleagues (2020) as our preliminary deductive coding framework. This framework was developed through a review of reviews on key domains of research partnerships and identifies the principles, strategies, outcomes and impacts reported in different types of research partnership approaches. Principles and strategies were defined as “how research partnerships work” and included approaches, facilitators, characteristics and mechanisms of partnerships. The overarching principles and strategies identified by Hoekstra and colleagues are described below:

| Subcategory                                       | Description                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                   |
|---------------------------------------------------|-----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| <b><i>Principles</i></b>                          |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                               |
| Relationship between researchers and stakeholders | Partners build and maintain relationships based on trust, credibility, respect, dignity, and transparency<br>Partners acknowledge, reward and value the diverse expertise of the partnership and its members<br>Partners share in decision-making and leadership of different research activities<br>The partnership addresses power dynamics within the team and aim to promote equity, self-determination and/or social justice<br>The partnership ensures representation and/or inclusivity and addresses disciplinary and sectoral issues |
| Co-production of knowledge                        | Partners co-produce knowledge and meaningfully engage stakeholders at different phases of the research process<br>Partners ensure that all members of the partnership have ownership over the data and resulting knowledge products<br>Partners strive to balance the need for scientific rigour alongside the practical need for actionable knowledge<br>Partners ensure the long-term implementation of the findings in real world settings and systems                                                                                     |
| Meaningful stakeholder engagement                 | Partners carefully plan and regularly reflect on their strategic approach to collaboration<br>Partners are flexible and creative in the collaborative research activities and tailor the approach<br>Researchers and stakeholders benefit from the partnership<br>The partnership identifies the stakeholder’s needs and makes sure that the research is relevant for the stakeholders                                                                                                                                                        |
| Capacity-building, support and resources          | Partners build capacity among all members of the partnership<br>Partners ensure bidirectional exchange of skills, knowledge and capacity between members of the partnership                                                                                                                                                                                                                                                                                                                                                                   |
| Communication between researchers                 | The partnership fosters regular, open, clear and honest communication between its members                                                                                                                                                                                                                                                                                                                                                                                                                                                     |

|                                                                         |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
|-------------------------------------------------------------------------|----------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------|
| and stakeholders                                                        |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
| Ethical issues of collaborative research activities                     | Ethical issues of collaborative research activities                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                              |
| <b>Strategies</b>                                                       |                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
| Relationship between researchers and stakeholders                       | <p>Initiate partnership and identify the team members; the partnership can be initiated by researchers or stakeholders; researchers can use targeted or open strategies to identify the stakeholders</p> <p>Monitor, experiment with and evaluate the collaborative research activities on an ongoing basis</p> <p>Work together to develop and define norms, rules and expectations in terms of timelines and tasks; this includes defining the level of stakeholders' engagement, roles and commitment</p> <p>Use a variety of activities to foster collaboration, communication and respect amongst the team members; strategies can include, but are not limited to, creating a common language, negotiating and addressing conflict, tailoring meets to the needs of the team, and providing opportunities to socialise</p> |
| Capacity-building, support and resources                                | <p>Provide opportunities to educate and train all team members; this strategy may include training that supports capacity for collaboration or research methods</p> <p>Provide time, resources and funding to support the collaborative research activities; stakeholders may be paid for engagement in the research process</p> <p>Provide practical and emotional support to stakeholders to help overcome barriers to engagement</p>                                                                                                                                                                                                                                                                                                                                                                                          |
| Communication between researchers and stakeholders                      | <p>Use a variety of methods to facilitate communication amongst team members; strategies include, but are not limited to, verbal methods (e.g. structured meetings, brainstorm sessions), written methods (e.g. email discussions, surveys) and visual methods (e.g. photovoice); this communication can be done in-person or via mediated methods (e.g. teleconference, online)</p>                                                                                                                                                                                                                                                                                                                                                                                                                                             |
| Stakeholder engagement in the planning of the research                  | <p>Strategies include, but are not limited to, stakeholder engagement in identifying or refining the 'research questions', stakeholder engagement in development the 'research protocol', stakeholder engagement developing or refining 'research instruments' (e.g. questionnaires, interview guides) and stakeholder engagement in development of participant 'information material' (e.g. informed consent)</p>                                                                                                                                                                                                                                                                                                                                                                                                               |
| Stakeholder engagement in conducting the research                       | <p>Strategies include, but are not limited to, stakeholder engagement in 'data collection' (e.g. recruitment of participants, study outcomes, conducting interviews, conducting literature review), stakeholder engagement in data analysis, and interpretation of findings</p>                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                                  |
| Stakeholder engagement in dissemination and application of the research | <p>Strategies include, but are not limited to, stakeholder engagement in 'writing reports or scientific papers' (e.g. stakeholder is co-author on a scientific paper), stakeholder engagement in 'presenting findings' to academic and community audiences, stakeholder engagement in a 'developing and implementation action plan' to ensure findings are used, and stakeholders use the findings to create change</p>                                                                                                                                                                                                                                                                                                                                                                                                          |

Two team members (O.M. & C.S.) led the analysis. Both researchers re-read the de-identified transcripts and familiarized themselves with the data. All transcripts were imported into Nvivo for analysis. The two team members applied the framework and coded transcripts using the principles

and strategies listed above as the initial labels, identifying anything that might be relevant from as many different perspectives as possible. After coding the first few transcripts, the two authors met to discuss the labels they applied to ensure consistency in the coding approach before coding all remaining transcripts. One author (O.M.) compared the data coded by both authors and developed the initial themes and identified illustrative quotations. These themes were then reviewed by all members of the research team who conducted interviews (L.L., J.K., A.L.) and revised based on feedback and their reflections on the interviews they conducted. Additional illustrative quotations were selected to highlight the perspectives of program managers, principal investigators, and payers of health research. All team members reviewed the final findings and interpretations, drawing on our multidisciplinary expertise in engaged research.

## Ethical Approval

This project received ethical approval from the Bruyere Research Institute (#M16-21-012), Ottawa, Canada. Approval letters and consent forms are available in Appendix 2 and 3, respectively.

## 5.4 RESULTS

A total of 43 interest-holders (65% female) from 15 different countries and all 10 interest-holder groups participated in the interviews (Table 2). Interest-holders had varied experiences in guideline development - some participants had substantial experience in leading guidelines, many had participated as members of guideline development groups, and some were guideline naive but had an interest in engaged research approaches.

**Table 2: Interview participant characteristics**

| Interest-holder group | No. interviews | Sex                | Countries                                             |
|-----------------------|----------------|--------------------|-------------------------------------------------------|
| Patients              | 7              | 5 Female<br>2 Male | Australia (n=1), Canada (n=2), UK (n=3),<br>USA (n=1) |

|                           |           |                               |                                                                                      |
|---------------------------|-----------|-------------------------------|--------------------------------------------------------------------------------------|
| Payers of health research | 1         | 1 Female                      | Canada (n=1)                                                                         |
| Payers of health services | 1         | 1 Female                      | Argentina (n=1)                                                                      |
| Peer Review Editors       | 8         | 2 Female<br>6 Male            | Argentina (n=1) Australia (n=1), Canada (n=1), Columbia (n=1), India (n=2), UK (n=2) |
| Polymakers                | 1         | 1 Female                      | Slovenia (n=1)                                                                       |
| Principal investigators   | 6         | 5 Female<br>1 Male            | Brazil (n=1), Canada (n=1), Columbia (n=1), Switzerland (n=1), USA (n=2)             |
| Product makers            | 2         | 2 Female                      | Canada (n=1), New Zealand (n=1)                                                      |
| Program managers          | 3         | 2 Female<br>1 Male            | Afghanistan (n=1), Canada (n=1), Estonia (n=1)                                       |
| Providers                 | 9         | 4 Female<br>5 Male            | Canada (n=3), Croatia (n=1), New Zealand (n=1), Nigeria (n=1), Peru (n=1), USA (n=2) |
| Public                    | 5         | 5 Female                      | Australia (n=1), Canada (n=2), USA (n=2)                                             |
| <b>TOTAL</b>              | <b>43</b> | <b>28 Female,<br/>15 Male</b> | <b>15 countries</b>                                                                  |

## Principles

***Shifting from a vertical power structure to a horizontal power structure.*** Vertical and horizontal power structures differ in how power is distributed and decision-making is organized. Vertical structures are hierarchical and centralized, while horizontal structures are more egalitarian. Traditionally, principal investigators and health care providers held positions of power throughout guideline development, leading the process and making the final decisions regarding health recommendations based on “the narrative of the clinical assessment of need”. Communication and information tend to flow from the top down, and feedback from others may be limited. Engaging diverse interest-holders demands a paradigm shift to a horizontal power structure, where power is distributed more evenly across individuals or groups, rather than being concentrated at the top of a power hierarchy. This structure emphasizes collaboration, equality, and shared decision-making throughout the guideline development process.



*“The root of all of this is a hierarchy, like a vertical power structure versus a horizontal power structure [...] Ideally, these [patients and healthcare providers] should be on an equal footing, my voice as a patient should have equal value to the healthcare professionals voice. Unfortunately, we still have some way to go to establish that... a lot of what we're talking about today is underpinned by power.”* (Patient 2, male, UK)

Several patients and members of the public emphasized the importance of being treated as a peer alongside healthcare providers and principal investigators and being provided with opportunities to make decisions together.

*“There isn't anything important that researchers should decide by themselves, any more than there's anything important about research, that patients should be deciding by themselves. We must start making these decisions together.”* (Patient 5, female, UK)

Providers agreed with this sentiment, emphasizing the importance of “some sort of paradigm shift to [...] shared decision making” (Provider 2, male, Nigeria). Members of the public highlighted that this shift in power towards shared decision making needs to be based on relationships built on trust, respect and “the belief of those people at the table [providers and principle investigators] that I should be there and I can contribute” (Public 3, female, Canada).

**Promotion of epistemic justice.** The term “epistemic justice” describes fairness and equality in the production, dissemination, and validation of knowledge. Epistemic justice emphasizes the recognition of diverse perspectives, experiences, and forms of knowledge, particularly those that have historically been marginalized or excluded. Co-production of knowledge was consistently described as an essential principle of guideline development to ensure that guideline recommendations address patient needs. One patient explained that there needs to be epistemic justice for patients and members of the public to share knowledge production spaces, “somewhere where we meet to establish common understandings, common narrative, shared learning” alongside clinicians in order to provide a complete knowledge base to inform guideline development.

*“Knowledge doesn't belong to anyone...any one group who thinks they've got all the knowledge they need is very delusional. [...] like a three legged stool, you can't have a two legged stool, you fall off. And if your two legs are academic knowledge and clinical knowledge, you're gonna keep falling off. So what you need is the knowledge of lived experience to stabilize you, then you've got a complete knowledge base. And that stands a chance of creating an evidence base that will lead to better care that will actually address all the needs of the clinicians and critically, the patients and people who care for them”* (Patient 5, female, UK)

Applying an epistemic justice approach to guideline development requires “embracing the not knowing” (Patient 5, female, UK) and encouraging bi-directional conversations that promote co-learning, where “we are going to learn from them [patients] as much as we're teaching them...maybe we'll learn from them more” (Principal investigator 4, female, USA). Importantly, one member of the public highlighted that patient experience is often viewed as “anecdotes and stories” (Public 2, female, Canada), and it does not receive the same weight and value as quantitative research. In order to embody epistemic justice in guideline development, knowledge based on experience needs to be valued as highly as traditional academic knowledge:

*“If you've got patients who've got very little kind of educational or so-called ‘academic ability’, but we've got extremely important lived experience that needs to be conveyed, then you need to find those places for them in the research world where they can provide that knowledge”* (Patient 5, female, UK)

**Representation.** Interest-holders generally recognized the importance of ensuring that diverse perspectives were represented within a guideline development group. However, how “representation” was described differed. For some interest-holders, representation meant that different groups, such as patients and providers of health care, were involved in decision making. For others, representation meant ensuring that geographic representation within a specific discipline was considered so that guideline recommendations would have local buy-in across a country. One provider of care emphasized the importance of including providers of care across

regional health authorities to ensure uptake, and paying special attention to Indigenous representation in their country context (Provider 8, male, New Zealand). However, a program manager explained that they have moved away from a speciality-based model, which focused on engaging people from specific clinical areas, towards an expertise-based model which could span several speciality areas:

*"So we've moved away from a specialty based model... we don't say 'you must be a college holding pediatric infectious disease specialist', we say the committee has to cover off these areas of expertise [...] it might be that we have a PhD epidemiologist who also is an infectious disease doctor, so they actually are satisfying the requirement to have expertise in multiple areas [...] that is outlined in the terms of reference, like it describes the content areas that are expected to be represented."* (Program manager 2, male, Canada)

Patient representation was discussed most in depth, primarily around concerns as to the ability of patients to represent the wider patient population:

*"But when is the person speaking from their own lived experience? And when are they claiming, rightly or wrongly, to represent cohorts of patients? I think a lot of people call themselves a representative, but then they speak from their own personal lived experience and are not really representing. Whereas when I claim to represent, that's because I've gone into my networks, and I've consulted hundreds of people [...] often I'll end up in a conversation with 100 or so people back and forth, back and forth, I say, what questions do you want me to ask? What information do you want me to pass on? And they'll tell me"* (Patient 3, female, Australia)

Additionally, principal investigators commented on the pragmatic decisions that guideline developers need to make, recognizing that it is not possible to have every patient, provider or program manager perspective represented in a single guideline project. They emphasized that, rather than “checking a box” by including individuals from several interest-holder groups, guideline developers should focus on including the perspectives that are essential for making the relevant guideline decisions, such as a patient “who had the major side effect from a certain intervention, or [...] who's not insured” (Principal investigator 4, female, USA)

**Avoiding tokenism and promoting diversity.** When engagement in guideline development is done tokenistically, a small number of individuals from underrepresented groups may be involved to give the appearance of diversity or inclusivity. Their presence may serve to deflect criticism or fulfill diversity quotas without meaningfully influencing the guideline initiative with their perspectives or experiences.

*“When you have an unknown health condition and you have a PhD kind of linked to health research, you get wheeled out and asked to participate in certain things for representation, I guess.” (Patient 2, male, UK)*

*“It's just that the way they went about it, they didn't bring in the lived experience people and the patient organization until after these academics had created the draft.” (Patient 3, female, Australia)*

Despite their inclusion, token individuals may have limited influence or decision-making power within guideline development. They may be excluded from important discussions or marginalized in decision-making processes.

*“I feel that I live in a PPI [patient and public involvement] cupboard. Every now and again, somebody needs some PPI. So they open the cupboard door, pull me and a few others out, sit us down and say this is what we want. This is what we're going to do. Is that alright with you? Okay, back in the cupboard, and we'll see you another time.” (Patient 5, female, UK)*

Overall, tokenism undermines genuine efforts to promote diversity, equity, and inclusion by reducing engagement to mere symbolic gestures rather than addressing systemic issues of exclusion. Patients recognized how this might exacerbate ongoing issues related to equity and diversity in guideline development. One patient (Patient 4, female, Canada) described most patient advocates as “a 50-something white woman” and explained that guideline developers need to work harder to engage young parents, older people, new immigrants and Indigenous

people. This was echoed by a second patient, who also mentioned that patient representatives are generally “white people that speak English and that have a high degree of education” (Patient 6, female, Canada).

## Strategies

***Governance and establishing committees.*** Patients recommended that a key strategy in ensuring their meaningful engagement in guideline development was involving them in governance alongside providers of care. Governance of guideline development includes overseeing the organization, budget and planning of the initiative. It also involves establishing clear roles, responsibilities, and reporting structures for project team members. Several patients believed that they should be involved in the governance of health guideline development.

*“I think that they should be involved in governance. Providers and patients and caregivers have the capacity and the skill and the relationships to be involved in making decisions from the beginning.”* (Patient 1, male, USA)

Another patient described that the “right” people for this role should have “*an appetite for change, who've got vision, who've got [...] the kind of emotional energy that's needed to bring others along with them.*” (Patient 5, female, UK). It was recognized, however, that having experience of a health condition would not be sufficient to fill this role - patients would also need to have strong language and scientific skills. Recruitment for these roles could be done through patient organizations and their social media channels, which are well connected to a broad range of patients in various locations.

If it is not possible to be involved in governance, one principal investigator (Principal investigator 6, female, Canada) suggested that some interest-holder groups including patients, policymakers and program managers can be involved in other committees or working groups to

allow them to engage in defined activities during guideline development. This includes acting as guideline development panel members, external review group members, and peer review group members. Interest-holders could also be engaged externally to the guideline development group, for example, as survey participants. It was noted that program managers, particularly those who may be too busy to engage regularly throughout the process, could be invited to participate in workshops at the end of the initiative:

*“If someone was at the provincial level, and it was not possible for them to be part of that [working group]. but it was very important to be engaged or provide inputs.... In this case, we just say to them, ‘Okay, when we complete the first draft, we will share with you that document and then we’ll have your point of view’ or also, [...] when we complete it, in order to have a wide range of engagement, we organize workshops at the national level, and invite all the key stakeholders to be part of that and having their inputs on that.”* (Program manager 1, female, Afghanistan)

In cases where there are insufficient resources to establish these committees or additional engagement activities, it was also recommended to look for published literature on interest-holder priorities:

*“I even say to a group, if you don't have the capacity to be having these extra committees and doing these large surveys and everything, then just look to the literature to what's already been done about stakeholder views, etc. That's something relatively easy without actually trying to find people and involve them and bring them in and have meetings with them. ... And, you know, they think, Oh, well, that's not really involving them. But it is, you're still getting information about their priorities”* (Principal investigator 6, female, Canada)

**Setting clear expectations and timelines.** Clear expectations ensure that everyone understands their roles and responsibilities within a guideline development initiative. One peer reviewed journal editor described this as “setting the boundaries and outlining your ask” (Peer reviewed journal editor 8, male, UK). When roles and timelines are well-defined, individuals can focus

their efforts on tasks that directly contribute to achieving objectives. Clear expectations contribute to interest-holder satisfaction by ensuring that deliverables are produced on time and meet the agreed-upon standards. This enhances trust and credibility with interest-holders, which is essential for maintaining positive relationships. When inviting interest-holders to join a guideline initiative, guideline developers should include:

*“a description of what's expected, how much of my time it is going to take, what is expected of me. So I always asked that question, what are you expecting? Are you expecting me to be writing these portions of the grant? Or are you expecting me to feed back some comments? [...] I think, if you understand clearly that there's this many meetings, like monthly meetings or quarterly meetings, and it's going to be a full day or it's two hours [...] those expectations should be clearly laid out before somebody gets involved. (Patient 6, female, Canada)*

Additionally, patients in particular expressed frustration with unclear or unrealistic timelines. This included timelines which were too short to adequately review documents and provide meaningful feedback, but also irritation with the long process of guideline development. One patient described that, while she had energy several years prior, the slow pace of academic work and her declining health meant that she had less energy to stay committed with projects: “for people who like a deadline, like, it's really slow” (Patient 4, female, Canada). This was echoed by a second patient:

*“I think it's even if the goodwill is there, and they could have been at the beginning of a year, the second year, their conditions deteriorate and they can't do it any longer. So I think those are sort of inequities that have got to be thought about and addressed.” (Patient 7, female, UK)*

To mitigate this frustration, regular follow-ups should be communicated to all interest-holders engaged in the guideline development project. One provider expressed the importance of “having clear agendas, having clear, regular meetings, making sure that the agendas are followed, and that decisions and actions are documented. Clearly, all of that kind of information, or all of that planning is really, really vital to success” (Provider 7, female, Canada).

**Compensation and acknowledgement.** Patient and public interest-holders may have different motivations and expectations regarding compensation compared to other interest-holders who engage in guideline development. For other interest-holders, participation in research or guideline activities is often an implied part of their job that helps to advance their career. In contrast, patients and members of the public often take time off of work, caregiving duties or other regular daily activities in order to contribute. Patients and members of the public expressed mixed feelings regarding compensation. Some patients reported turning down all compensation, others expressed that patients should be compensated for their out-of-pocket expenses but not for sharing their experiential knowledge, and others felt that offering compensation for the time spent towards a guideline was a good way to recognize patients' contributions and value. These differences in opinion may have been related to country context. For example, one Canadian patient explained that accepting financial compensation could influence a person's eligibility to receive the Canada Pension Plan disability benefit. In other contexts, such as the UK and Australia, patients explained that including budget items in grant applications to support consultation and co-design with patients was an expectation and improved chances of receiving funding. Engaging patients early in grant submissions was suggested as a way to plan for these costs:

*“I get really cross when in the budget and the planning and the organizing there are false assumptions made about what will be accessible and inclusive engagement with consumers, and what the costs and what the resources will be that are required to make it accessible and inclusive. So to me, I would want to be involved [early], or know that there's someone involved who understands our patients.” (Patient 1, male, USA)*

Another patient suggested that organizations or institutions who develop guidelines should have an organizational-level group or committee to assist in the pre-application grant process. This group could review grant submissions and indicate strategies to improve patient and public engagement, including providing suggestions for compensation. Patients also suggested that, in situations where financial compensation cannot be accepted, co-authorship on guideline documents could be a potential mechanism to recognize patient and public contributions:



*“It’s actually quite terrible. They put out a paper and they didn’t even put a thank you to the patients [...] I know some patients who’d spent hours looking through all the research questions that had been submitted. They didn’t even get the acknowledgement, let alone co-authorship, and they really should have been co author, and therefore as co author, they should have maybe seen the piece before it was published... a very bad example”* (Patient 4, female, Canada)

***Strategies to facilitate engagement throughout the guideline development process.*** Across all interest-holder groups, there was a clear preference to be engaged early in the guideline development initiative, particularly in setting priorities, topic selection and research question generation. Engagement in other aspects of guideline development was more variable depending on the technical skills required. Some strategies were identified to support interest-holder engagement in technical processes such as evidence reviews and developing recommendations:

1. *Conduct interest-holder mapping:* Program managers highlighted the importance of mapping exercises to identify and recruit the key players working in the field of the guideline. They emphasized that the individuals with the highest level of interest should be engaged to ensure retention throughout the guideline development project.
2. *Mentorship:* For patients and public members, a mentorship program was suggested whereby less experienced patients/public could “buddy up” with a more experienced patient/public member to guide them through the guideline development process to help their advocacy efforts be most effective.
3. *Facilitation by a methodologist or chair:* One principal investigator recognized that some interest-holders, such as program managers, may not be familiar with technical processes such as determining the strength of the evidence. They suggested that guideline group discussion includes a skilled methodologist or chair who could facilitate discussions about interpretation, such as the size of the benefits and harms.
4. *Leveraging technology and online meeting spaces:* Patients were generally supportive of online engagement opportunities because it allows patients from different geographic locations to participate in guideline development. However, one principal investigator cautioned that online meetings are only effective for small group discussions. When

larger groups need to meet, it can be difficult to ensure that everyone gets a chance to provide their input in an online environment. While online meetings can help reduce guideline development costs, it may still be necessary to invest in in-person meetings and travel expenses. They highlighted that online tools, such as GRADE Panel Voice, can help coordinate the voting processes.

5. *Meeting people “where they are, as opposed to asking them to come to you”*: One patient described a successful approach to engaging with other patients is meeting in familiar local spaces within the community rather than academic locations such as a university. They described the success of meeting with patients at the local pub, knowing that the patients lived nearby, parking was accessible, the food was enjoyable, and the patients knew where to locate an accessible washroom.

***Expanding opportunities for engagement in dissemination activities.*** Several interest-holders emphasized the importance of looking beyond academic publications as an opportunity for dissemination. Interest-holders should be empowered to plan and participate in dissemination activities:

*“People can also be empowered to present about this. So a lot of the time again, you see that only the chair has one session in one meeting, they present about a certain guideline, which is completely the wrong approach. You actually want to see that every person on that panel have an opportunity to showcase the work, because then they feel it's their responsibility to do this dissemination [...] it makes people feel like it's not ‘I'm just there and I'll get a publication out of it’. It's way bigger and more important.”* (Principal investigator 4, female, USA).

For some interest-holder groups, this may mean presenting at annual academic meetings that are specific to a clinical or public health speciality. However, other strategies were also suggested, such as developing online education sessions led by providers, or developing social media campaigns led by patients, caregivers and members of the public. Planning for these dissemination activities should occur early and be re-visited regularly during the guideline

development process. One payer of health research explained that maintaining close relationships with implementing organizations can facilitate dissemination activities:

*“And so then we already have that sort of dissemination implementation mechanism [...] because we have that standing, ongoing connection with the provinces and territories. Other than that, as soon as we release new guidance we have a whole sort of media stream that we work with, to get the information out there just to the general public.”* (Payer of health research 1, female, Canada)

**Consider when engagement may not be feasible.** While most interest-holders recognized the importance of engagement in guideline development, they also highlighted several circumstances where engagement might be limited or not be possible. For example, in emergency contexts where rapid guidance is required, such as during the COVID-19 pandemic, it was recognized that only a subset of interest-holders (such as providers of care) would likely be engaged, and that guideline developers would need to make pragmatic decisions about the most essential interest-holder groups to engage in decision-making.

Additionally, two providers of care discussed that some countries, such as low-and-middle income countries or smaller countries, may not have capacity for full de novo guideline development, but rather may rely on guideline adaptation to inform their own national guidelines because they lack *“the sort of logistics in terms of funding, and [...] the appropriate capacity to sit in the panels”* (Provider 2, male, Nigeria). However, they did describe that it could be possible to engage with interest-holders who may not have been engaged in the original guidelines as part of the adaptation process to enrich the perspectives considered within their own national guideline documents.

## 5.5 DISCUSSION

The findings of this paper complement ongoing work by the MuSE Consortium to develop guidance for the engagement of interest-holders in guideline development (Petkovic et

al., 2020). Through interviews, we identified several principles and strategies to promote engagement across 10 different groups of interest-holders. The principles we identified were establishing horizontal power structures, promoting epistemic justice and focusing on co-production of knowledge, conceptualizing “representation” both across and within interest-holder groups, as well as avoiding tokenism and promoting diversity. Effective strategies for meaningful engagement included involving interest-holders in governance, setting clear expectations and timelines, providing compensation and acknowledgment for contributions, and providing opportunities for engagement in dissemination activities. We also identified specific strategies to facilitate engagement throughout the guideline development process. In addition, we highlight instances where engagement may not be feasible, including in emergencies where rapid guidance is required.

The findings of this research are unique in that the principles and strategies for engagement come directly from interest-holders themselves. Previous research in the area of principles and strategies for engaged guideline development has relied on published literature (e.g., Pinho-Gomes 2022; Petkovic et al., submitted; Magwood et al., submitted) and was lacking in evidence across many interest-holder groups. However, some interest-holder groups (such as patients, public, providers of care, and principal investigators) are oversampled among our interview participants. While these may represent the most commonly engaged groups in guideline development (Petkovic et al., submitted), this also means that our knowledge of the principles and strategies of some groups of interest-holders remain limited, as we did not achieve data saturation for each interest-holder group. Despite this, many principles and strategies identified in this study were consistent across interest-holder participants, suggesting that there are some core principles which apply to guideline development regardless of interest-holder group. In addition, we believe that many of the identified principles and strategies also apply outside the context of guideline development (e.g., in health research more generally).

Representation remains a key issue for interest-holder engagement in the development of guidelines (Petkovic et al., 2023), and this was echoed by interview participants. Current guidance on identifying interest-holders from the MuSE Consortium highlights that engaged individuals should be able and willing to represent an interest-holder group, but does not provide

concrete steps on how to do so (Parker et al., 2022). Smith et al. (2009) proposed a representation framework to serve as a guide for recruitment of interest-holders which incorporates four elements: ‘nominated membership’ (members who are nominated by their organizations), ‘dispositional representation’ (individuals are engaged by virtue of their job role), representation of ‘shared interest’ (advocates speaking on behalf of a group) and ‘personal representation’ (individuals with personal experience of the condition of interest). However, they also highlighted a pragmatic consideration for interest-holder selection: how informed or skilled interest-holders need to be in relation to the task at hand (Smith et al., 2009). Indeed, it is unlikely that the average patient or healthcare provider, for example, has the necessary interest and experience to fully engage in health guideline development. Further research on how to recruit interest-holders and balance their representativeness and expertise within the context of guideline development is warranted.

Although principles are not “how to” guides, they should clearly identify what is desirable and ideal and be linked to action so that guidance can be developed based on those foundational principles (Pinho-Gomes et al., 2022). The current study informs the MuSE Consortium’s guidance for engagement in guideline development (Petkovic et al., 2020; Petkovic et al. submitted, Magwood et al., in progress). These findings also complement other guidance available from the MuSE Consortium to guide engaged research approaches, including practical guidance for involving interest-holders in health research (Concannon et al., 2019), factors to consider during identification and invitation of individuals in a research partnership (Parker et al., 2022), and a new taxonomy for defining conflicting and non-conflicting (legitimate) interests of interest-holders in health research (Akl et al., 2023). The MuSE Consortium has also begun a project to further explore the engagement of interest-holders in evidence synthesis (Tugwell et al., 2023).

### **Strengths and limitations**

While we interviewed ten different groups of interest-holders, recruitment across groups was unbalanced. We experienced challenges in recruiting several groups including program managers, policymakers, payers of health research and health services, and product makers. This may be reflective of the disproportionate membership of the MuSE Consortium itself (which has,

to date, mostly attracted principal investigators, providers of care, and patients/public members). This may also reflect these groups (un)willingness to participate in interview-based research or limited experience in guideline development. Our project was also constrained by its timing during the COVID-19 pandemic, when many program managers or policymakers may have had competing priorities (e.g., developing rapid guidance for COVID-19, vaccination implementation, etc). Additionally, while we aimed to recruit interviewees globally, participants (particularly patients and members of the public) were predominantly from high-income English-speaking countries. This generally reflects current practices in engaged health research (Jull et al., 2019), and more effort is needed to offer opportunities and build capacity in low- and middle-income countries.

## **5.6 CONCLUSIONS**

Engagement is time consuming and resource intensive and requires an important shift in power structures and governance. Guideline developers should do their best to integrate interest-holders in committees, engage them as early as possible in the process, provide them with rightful acknowledgment, be clear about expectations and timelines, and facilitate their engagement throughout the guideline development process. Guideline developers may need to make pragmatic decisions as to who they engage in guideline development and how, including at which stage of the process, and with what decision-making power. Constraints may exist when rapid guidance is needed or in settings with poor capacity for evidence synthesis and guideline development. Capacity strengthening in low- and middle-income countries may help address current disparities in engagement in guideline development.

Future research should explore issues around representativeness of interest-holders and testing of our proposed strategies. Additional investigation on the perspectives of program managers, policymakers, and payers of health research and health services would enhance the current findings.

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## **APPENDIX 1: INTERVIEW GUIDE (Patients)**

### **Section 1: Getting to know you**

First, I'd like to know a little about you. Could you please tell me about yourself?

- *E.g. where do you live, do you work, do you also have other roles (e.g. researcher, editor, program manager) etc.*

Thank you. Next, we would like to get your opinion about how and when stakeholders should be engaged in the guideline development process.

When we say 'guidelines' we are referring to clinical practice guidelines, which guide decision making about treatments, as well as public health guidelines, such as those you're probably familiar with related to COVID-19, like wearing a mask when in public.

When thinking about guidelines, which term do you prefer to describe yourself:

- 1) Patient
- 2) Consumer
- 3) Member of the public
- 4) Other?

Are there differences between these terms? If yes, what are they?

Could you please tell me about your experience in health guideline development?

- *Have you ever been involved in a guideline panel?*
- *How many years of experience do you have in this area? / How many guidelines?*
- *Do you remember the type of guideline you were a part of?*
- *What was your role in the guideline development process?*

## **Section 2: GIN-McMaster checklist**

Last year we surveyed stakeholders from around the world. They gave us their opinions on when they should be engaged in guideline development. At each stage, they had the choice of selecting “engagement not required”, advice/feedback, or decision-making.

Advice/feedback means that the stakeholder gives their opinions, and the guideline development team gets to decide whether they want to use those opinions.

Having a decision-making role means that stakeholder want an equal “vote at the table” – the guideline team *must* take their vote under consideration.

Now, I would like to show you the 18 stages of the GIN-McMaster checklist that patients have provided us feedback on how they want to be involved.

In the table on the screen, you can see that there are some stages where we have heard consistent opinions about engagement. In other stages, we are still unclear/unsure about how patients/public want to be engaged

We want to know

- 1) Do you agree with level of engagement indicated at each stage?
- 2) What are your opinions about the stages marked as “unclear”?

### **For Patients/Caregivers:**

| <b>GIN-MCMaster CHECKLIST</b>                                                              | <b>Engagement</b> |
|--------------------------------------------------------------------------------------------|-------------------|
| 1. Organisation, budget, planning                                                          | unclear           |
| 2. Priority Setting                                                                        | Decision-making   |
| 3. Guideline group membership                                                              | Decision-making   |
| 4. Guideline group processes                                                               | Decision-making   |
| 5. Identifying Target Audience and Topic Selection                                         | unclear           |
| 6. Consumer and Stakeholder Involvement                                                    | Decision-making   |
| 7. Conflict of Interest Considerations                                                     | unclear           |
| 8. (PICO) Question Generation                                                              | Decision-making   |
| 9. Considering Importance of Outcomes and Interventions, Values, Preferences and Utilities | Decision-making   |

|                                                                                                |                 |
|------------------------------------------------------------------------------------------------|-----------------|
| 10. Deciding what Evidence to Include and Searching for Evidence                               | unclear         |
| 11. Summarizing Evidence and Considering Additional Information                                | unclear         |
| 12. Judging Quality, Strength or Certainty of a Body of Evidence                               | unclear         |
| 13. Developing Recommendations and Determining their Strength                                  | unclear         |
| 14. Wording of Recommendations and of Considerations of Implementation, Feasibility and Equity | Decision-making |
| 15. Reporting and Peer Review                                                                  | Decision-making |
| 16. Dissemination and Implementation                                                           | Decision-making |
| 17. Evaluation and Use                                                                         | unclear         |
| 18. Updating                                                                                   | unclear         |

### **Section 3: Scenario**

Imagine that a new clinical practice guideline is being development for the **treatment of COVID-19 (e.g. a new drug)**. How and when do you think patients/caregivers/public should be engaged?

### **Section 4: Final remarks**

- If you could give one piece of advice to a guideline development team on how to engage with patients/public, what advice would you give to them?
- Please tell me anything you think we should know about engagement in guideline development.
- Is there anything else you would like to share?
- Do you have any questions for me?

Thank you [end interview].

## APPENDIX 2: ETHICS APPROVAL LETTER

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Centre de médecine familiale Bruyère  
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Tél./Tel.: 613-241-3344  
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Centre de médecine familiale Primrose  
Primrose Family Medicine Centre  
35, rue Primrose St.

Thursday, August 19, 2021

**Dr. Peter Tugwell**

**Re:** "Multi-Stakeholder Engagement in Guidelines Development"  
(Bruyère REB Protocol #M16-21-012)

### **Amendment Approval**

**Amendment dated:** June 17, 2021

Dear Dr. Tugwell,

Thank you for submitting an amendment request for the above named project.

The following has been approved:

- ✓ BREB, version date: August 7, 2021;
- ✓ Email Invitation, version date: August 14, 2021;
- ✓ Interview Guide, version date: July 14, 2021;
- ✓ ICF Verbal, version date: August 14, 2021;
- ✓ ICF, version date: August 19, 2021

The Bruyère Continuing Care REB complies with the membership requirements and operates in compliance with the Tri-Council Policy Statement 2: Ethics Conduct for Research Involving Humans; the International Conference on Harmonization - Good Clinical Practice: Consolidated Guideline; the provisions of the Personal Health Information Protection Act 2004; and the Food and Drug Act of Health Canada and its applicable Regulations.

The information received has been reviewed and I hereby give approval on behalf of the REB.

Sincerely,

## APPENDIX 3: CONSENT FORM



### RESEARCH INFORMED CONSENT FORM

**Project Title:** Multi-Stakeholder Engagement in Guidelines Development

**Version Date:** August 18, 2021

#### Name and Contact Information of Researchers

Dr. Peter Tugwell, University of Ottawa

Email: XXXl@uottawa.ca

Supervisor and Contact Information: *Jennifer Petkovic, XXX@uottawa.ca*

**Project Sponsor and Funder (if any):** This project is funded by the Canadian Institute for Health Research (CIHR).

#### Bruyère Research Ethics Board Approval

Date of Approval: October 2021

Study # M16-21-012

#### Invitation

You are invited to take part in a follow-up interview for the Multi-Stakeholder Engagement in Guidelines Development (MuSE) project because you previously expressed interest in sharing your thoughts. The information in this form is intended to help you understand what we are asking of you so that you can decide whether you agree to participate in this study. Your participation is entirely voluntary, and a decision not to participate will not be used against you

in any way. As you read this form, and decide whether to participate, please ask all the questions you might have, take whatever time you need, and consult with others as you wish.

### **What is the purpose of the study?**

This study aims to help guideline developers by providing guidance on how and when to involve stakeholders in the guideline development process. We are doing this by asking stakeholders (e.g. patients), like yourself, what is important to them and how they want to be considered when guidelines are being made so that the recommendations that are made will reflect health outcomes patients believe are important. We will use what we find to develop a list of recommendations to include in a guidance checklist for guideline developers.

### **What will I be asked to do?**

The study involves an interview to explore your views on why your stakeholder group should be engaged in some specific steps as decision makers and how that engagement might look like in practice. The interviews will occur virtually using videoconferencing software (Zoom).

With your consent, this interview will be audio and video-recorded, and transcribed by a member of our team. The interview files will be encrypted and stored in a secure cloud platform (Zoom US). Once transcribed, the recording will be retained for 7 years prior to being destroyed. Your data will be anonymized.

We expect that this interview will take about 25 to 40 minutes to complete. Your participation is voluntary, and you may choose not to take part, or not to answer any of the questions we ask.

### **Risks and Inconveniences**

We do not anticipate any risks to participating in this study.

### **Possible Benefits**

You may not receive any direct benefit from your participation in this study. However, your participation may allow researchers to improve how patients participate and engage with other stakeholders in the guideline development process

### **Compensation/Incentives**

You will not be paid or compensated for your participation in this study.

### **No waiver of your rights**

By signing this form, you are not waiving any rights or releasing the researchers from any liability.

### **Withdrawing from the study**

If you withdraw your consent during the course of the study, all information collected from you before your withdrawal will be discarded.

You may request that your data be removed from the study and deleted by notice given to the Principal Investigator (named above) within one week of the interview.

### **Confidentiality**

We will remove all identifying information from the study data as soon as possible, which will be after we transcribe and verify the interview data. We will treat your personal information as confidential, although absolute privacy cannot be guaranteed. However, research records identifying you may be accessed by the Bruyère Continuing Care Research Ethics Board for the purpose of monitoring the research.

The results of this study may be published or presented at an academic conference or meeting, but the data will be presented so that it will not be possible to identify any participants unless you give your express consent. De-identified data from this study may be shared with other researchers for verification, and to permit them to build upon our findings.

You will be assigned a code so that your identity will not be directly associated with the data you have provided. Your data will be stored and protected by Zoom US in a server located in the USA but may be disclosed via a court order or data breach.

We will encrypt any research data that we store or transfer.

### **Data Retention**

After the study is completed, your de-identified data will be retained for a period of 7 years and then securely destroyed.

### **New information during the study**

In the event that any changes could affect your decision to continue participating in this study, you will be promptly informed.

### **Ethics review**

This project has been reviewed and approved by the Bruyère Continuing Care Research Ethics Board as study #M16-21-012. If you have any ethical concerns about the study, or the way it is conducted, please contact the Bruyère Continuing Care REB: (613) 562-6262 Ext. 4003. Please note that during the time of the COVID-19 pandemic it is usually not possible to reach the REB staff by telephone. Accordingly, until more normal times, please contact the REB by email at [REB@bruyere.org](mailto:REB@bruyere.org)

### **Statement of consent – print and sign name**

I \_\_\_\_ [*name of study participant*]\_\_\_\_\_, have read the information given in this informed consent and all my questions have been answered to my satisfaction. I have had sufficient time to consider whether to participate in this study. I understand that my participation in this study is voluntary and that I may withdraw from the study at any time without penalty.



I voluntarily agree to participate in this study. Yes \_\_\_\_\_ No \_\_\_\_\_

I agree to be audio recorded. Yes \_\_\_\_\_ No \_\_\_\_\_

I agree to be video recorded Yes \_\_\_\_\_ No \_\_\_\_\_

I would like you to send me a summary of results from this study when they are available.

\_\_\_\_ Yes \_\_\_\_ No      Email: \_\_\_\_\_

\_\_\_\_\_

Signature of participant

Date:

\_\_\_\_\_

Team member who interacted with the participant

Date:

To the best of my knowledge, the information in this consent form, and the information that I, *(print name)* \_\_\_\_\_ have provided in response to any questions, fairly represents the project. I am committed to conducting this study in compliance with all the ethical standards that apply to projects that involve human subjects. I will ensure that the subject receives a copy of this consent form.

\_\_\_\_\_

Signature of researcher

\_\_\_\_\_

Date

## Consolidated criteria for reporting qualitative studies (COREQ): 32-item checklist

| No                                             | Item                                     | Guide questions/description                                                                                                                      | Page    |
|------------------------------------------------|------------------------------------------|--------------------------------------------------------------------------------------------------------------------------------------------------|---------|
| <b>Domain 1: Research team and reflexivity</b> |                                          |                                                                                                                                                  |         |
| Personal Characteristics                       |                                          |                                                                                                                                                  |         |
| 1.                                             | Interviewer/facilitator                  | Which author/s conducted the interview or focus group?                                                                                           | 313-314 |
| 2.                                             | Credentials                              | What were the researcher's credentials? <i>E.g. PhD, MD</i>                                                                                      | 313-314 |
| 3.                                             | Occupation                               | What was their occupation at the time of the study?                                                                                              | 313-314 |
| 4.                                             | Gender                                   | Was the researcher male or female?                                                                                                               | 313-314 |
| 5.                                             | Experience and training                  | What experience or training did the researcher have?                                                                                             | 313-314 |
| Relationship with participants                 |                                          |                                                                                                                                                  |         |
| 6.                                             | Relationship established                 | Was a relationship established prior to study commencement?                                                                                      | 313-314 |
| 7.                                             | Participant knowledge of the interviewer | What did the participants know about the researcher? <i>e.g. personal goals, reasons for doing the research</i>                                  | 313-314 |
| 8.                                             | Interviewer characteristics              | What characteristics were reported about the interviewer/facilitator? <i>e.g. Bias, assumptions, reasons and interests in the research topic</i> | 313-314 |
| <b>Domain 2: study design</b>                  |                                          |                                                                                                                                                  |         |
| Theoretical framework                          |                                          |                                                                                                                                                  |         |
| 9.                                             | Methodological orientation and Theory    | What methodological orientation was stated to underpin the study? <i>e.g. grounded theory, discourse analysis,</i>                               | 314-317 |

| No                                                  | Item                         | Guide questions/description                                                               | Page       |
|-----------------------------------------------------|------------------------------|-------------------------------------------------------------------------------------------|------------|
| <i>ethnography, phenomenology, content analysis</i> |                              |                                                                                           |            |
| Participant selection                               |                              |                                                                                           |            |
| 10.                                                 | Sampling                     | How were participants selected? <i>e.g. purposive, convenience, consecutive, snowball</i> | 314        |
| 11.                                                 | Method of approach           | How were participants approached? <i>e.g. face-to-face, telephone, mail, email</i>        | 314        |
| 12.                                                 | Sample size                  | How many participants were in the study?                                                  | 318-319    |
| 13.                                                 | Non-participation            | How many people refused to participate or dropped out? Reasons?                           | NR         |
| Setting                                             |                              |                                                                                           |            |
| 14.                                                 | Setting of data collection   | Where was the data collected? <i>e.g. home, clinic, workplace</i>                         | 314-315    |
| 15.                                                 | Presence of non-participants | Was anyone else present besides the participants and researchers?                         | N/A        |
| 16.                                                 | Description of sample        | What are the important characteristics of the sample? <i>e.g. demographic data, date</i>  | 318-319    |
| Data collection                                     |                              |                                                                                           |            |
| 17.                                                 | Interview guide              | Were questions, prompts, guides provided by the authors? Was it pilot tested?             | Appendix 1 |
| 18.                                                 | Repeat interviews            | Were repeat interviews carried out? If yes, how many?                                     | N/A        |

| No                                     | Item                           | Guide questions/description                                                                                                                 | Page    |
|----------------------------------------|--------------------------------|---------------------------------------------------------------------------------------------------------------------------------------------|---------|
| 19.                                    | Audio/visual recording         | Did the research use audio or visual recording to collect the data?                                                                         | 315     |
| 20.                                    | Field notes                    | Were field notes made during and/or after the interview or focus group?                                                                     | N/A     |
| 21.                                    | Duration                       | What was the duration of the interviews or focus group?                                                                                     | 315     |
| 22.                                    | Data saturation                | Was data saturation discussed?                                                                                                              | 331     |
| 23.                                    | Transcripts returned           | Were transcripts returned to participants for comment and/or correction?                                                                    | N/A     |
| <b>Domain 3: analysis and findings</b> |                                |                                                                                                                                             |         |
| Data analysis                          |                                |                                                                                                                                             |         |
| 24.                                    | Number of data coders          | How many data coders coded the data?                                                                                                        | 317     |
| 25.                                    | Description of the coding tree | Did authors provide a description of the coding tree?                                                                                       | 316-317 |
| 26.                                    | Derivation of themes           | Were themes identified in advance or derived from the data?                                                                                 | 316-318 |
| 27.                                    | Software                       | What software, if applicable, was used to manage the data?                                                                                  | 317     |
| 28.                                    | Participant checking           | Did participants provide feedback on the findings?                                                                                          | N/A     |
| Reporting                              |                                |                                                                                                                                             |         |
| 29.                                    | Quotations presented           | Were participant quotations presented to illustrate the themes / findings?<br>Was each quotation identified? e.g. <i>participant number</i> | 318-330 |

| No  | Item                         | Guide questions/description                                            | Page    |
|-----|------------------------------|------------------------------------------------------------------------|---------|
| 30. | Data and findings consistent | Was there consistency between the data presented and the findings?     | 318-330 |
| 31. | Clarity of major themes      | Were major themes clearly presented in the findings?                   | 318-330 |
| 32. | Clarity of minor themes      | Is there a description of diverse cases or discussion of minor themes? | NR      |

## **CHAPTER 6: DISCUSSION**

### **Chapter Overview**

This Chapter summarizes study findings from all three articles included in this thesis. Additionally, it discusses three main key considerations identified from these articles: issues regarding representation, reporting of equity considerations, and common principles to support engagement. It concludes by discussing implications for population health practice and future research.

### **6.1 Summary of findings**

This thesis examines approaches for interest-holder engagement in evidence synthesis and health guideline development. Its goal is to inform the guidance developed by the MuSE Consortium's equity-oriented guidance for use by evidence synthesis researchers and guideline developers for the engagement of 11 different groups of interest-holders. The articles included in this thesis address knowledge gaps related to equity considerations, barriers and facilitators, and principles and strategies for engagement of interest-holders in evidence synthesis and health guideline development.

#### **6.1.1 Summary of Article 1**

Our scoping review of interest-holder engagement in evidence synthesis critically examined definitions and terminology. The term “stakeholder” was the most common umbrella term used to describe an individual or group who should be engaged in evidence synthesis. To describe engagement, the terms “involvement” and “engagement” were used interchangeably, often qualified as “meaningful” engagement. No included articles specified terms related to “equitable engagement”, such as “equity-oriented” or “equity-centered”. Few included publications described the interest-holders they engaged according to the PROGRESS-Plus mnemonic, making it difficult to determine considerations for engaging with diverse populations. Based on our scoping review findings, interest-holder engagement in evidence synthesis is concentrated in high-income English-speaking countries. Issues related to access to technology can result in resource bias and exclude people from lower socio-economic positions. We identified

considerations for engaging with youth and people living with disabilities, such as providing tailored training and support. Further, we identified four principles to support equitable engagement: Addressing power dynamics, accessibility, promoting diversity and inclusion, and ethical considerations. While these findings were identified from literature specific to evidence synthesis, these principles are applicable to engagement across evidence ecosystems, including primary research and health guideline development.

### **6.1.2 Summary of Article 2**

In our qualitative evidence synthesis, we identified a total of 52 findings encompassing 24 barriers and 28 facilitators to interest-holder engagement in health guideline development. While the majority of reported evidence was about patient engagement, many findings are applicable to all interest-holder groups and should be considered by guideline developers who aim to apply an engagement approach. Common barriers included a lack of organizational resources to support engagement activities, issues selecting representative interest-holders, unclear roles and responsibilities, power imbalances, tokenism, and a lack of technical skills.

By implementing an organizational engagement model, guideline developers can ensure that they have the necessary human and financial resources in place to support interest-holders across diverse perspectives, roles, and personal characteristics. Practical, technical and emotional support includes providing training to empower interest-holders and improve self-efficacy and confidence in guideline development skills, including evidence synthesis. Engagement throughout all stages of guideline development supports more meaningful engagement compared to occasional involvement. A skilled chair can manage group dynamics and support the contributions of all those involved in guideline development.

### **6.1.3 Summary of Article 3**

When we asked 43 interest-holders how to best support their engagement in guideline development, interview participants described principles such as establishing horizontal power structures, promoting epistemic justice, focusing on co-production of knowledge,

conceptualizing “representation” both across and within interest-holder groups, as well as avoiding tokenism and promoting diversity. Effective strategies for meaningful engagement included involving interest-holders in governance, setting clear expectations and timelines, providing compensation and acknowledgment for contributions, and providing opportunities for engagement in dissemination activities. We also identified specific strategies to facilitate engagement throughout the guideline development process. These included structured mapping exercises, mentorship, facilitation by a methodologist or chair, and leveraging technology.

In all, the articles included in this thesis contribute towards the body of work being conducted by the MuSE Consortium to develop guidance for engagement in health evidence syntheses and guideline development. When complemented by other articles within the MuSE research program (see Chapter 2, Figure 1), these articles offer comprehensive evidence to support the operationalization of the upcoming MuSE guidance for engagement in health guideline development (Petkovic et al., submitted; Magwood et al., in progress; Petkovic et al., 2020) and evidence synthesis (Tugwell et al., 2023).

## **6.2 Representation: A key issue in evidence ecosystems**

Evidence ecosystems rely on the engagement of diverse interest-holders to inform health decision-making (Gough et al., 2019). Appropriate representation of interest-holders can influence how evidence is interpreted and incorporated into policy and practice (Sohn, 2018). Decisions that consider the perspectives and needs of all interest-holders are more likely to balance economic, social, and environmental considerations. When interest-holders are adequately represented, the decision-making process gains legitimacy (Arnesen & Peters, 2017). A common theme across all three articles included in this thesis is around “representation” and what it means to be a “representative” for an interest-holder group. The MuSE Consortium previously identified representation as a key issue for engaged guideline development (Petkovic et al., 2023), and these findings were also highlighted in the literature and through interviews with interest-holders. “Representation” in health research is often contested and can describe whether an individual is “acting for” a group, such as defending their interests or being nominated as a representative, or “standing for” a group, such as sharing a particular experience



or being statistically similar to the represented group (Maguire & Britten, 2017). But how do we ensure that the interest-holders that engage with us are “representative” of the wider interest-holder group and that they are able to speak on behalf of a group rather than their own opinions and experiences? Our qualitative evidence synthesis, and additional literature (e.g., Sipila et al., 2022) found there can be significant challenges to recruit appropriate people from target groups that are capable of representing the wider population. The range of different definitions of what it means to represent, or to be a representative, helps to explain why “representation” in health research is so easy to challenge (Maguire & Britten, 2017). Particular issues for evidence synthesis and guideline development include representation across geographic contexts, socio-economic positions, educational experiences or cultural, linguistic, or gender identities. More guidance and research is needed on how to identify and select interest-holder representatives, and decide which diversity characteristics are most important to ensure appropriate representation for the decision-making context.

### **6.3 Equity considerations and characteristics are seldom reported**

Integrating health equity into evidence synthesis, guideline development and broader health decision-making requires the engagement of interest-holders, particularly representatives of disadvantaged population groups, who understand how to take health equity, human rights, gender and social determinants into account (Shaver et al., 2023). This promotes social justice by ensuring that these marginalized or excluded groups have a voice in processes that affect their lives. Unfortunately, the concept of “equitable engagement” in the evidence synthesis literature remains in its infancy, despite the movement towards equity-centered engagement in health more broadly (e.g., the *Supporting equity-centred engagement guide* (Ul Haq, 2023), the *Equity in engagement framework* (HQO, 2020), and PCORI’s *Equity and inclusion guiding engagement principles* (PCORI, 2021)). Additionally, the term “stakeholder” remains consistently used within evidence ecosystems to refer to individuals or groups who are responsible for or affected by health decision-making. No publications included in our scoping review on equity considerations for engagement in evidence synthesis acknowledged the colonial origins of the term nor did they recognize how continued use of the term perpetuates inequity. Critically reflecting on terminology and working towards decolonization are essential to population health

(Hayward et al., 2021). Language and terminology often reflect the power structures within a society, and many terms carry implicit assumptions and prejudices that perpetuate inequity and marginalization. By examining and revising the term “stakeholder” to “interest-holder” we can promote inclusivity, respect cultural identities, and ensure that diverse perspectives are valued in decision-making. Decolonizing our language challenges Eurocentric viewpoints and also advances social justice by addressing and reconciling systemic marginalization.

Our scoping review and qualitative evidence synthesis both identified limited reporting of personal characteristics of engaged interest-holders by PROGRESS or other dimensions of equity. On the one hand, this may be a deliberate choice by authors in order to protect the privacy of the interest-holders they work with. This could also represent a lack of guidance on reporting of equity, positionality or reflexivity in health research (Secules et al., 2021). While guidance exists for reporting equity in randomized controlled trials (CONSORT-Equity; Welch et al., 2017) and there is upcoming guidance for epidemiological observational studies (STROBE-Equity; Funnell et al., 2023), there is not much guidance for other study designs. Much of our evidence is extracted from qualitative studies, retrospective case reports or commentaries outlining author experiences. From what is reported, engaged evidence synthesis research and guideline development is concentrated in high-income settings among and English-speaking interest-holders, and very little is known about engaging interest-holders from low-income settings, diverse cultures or languages, or different socio-economic positions. This suggests a need for capacity building in low- and middle-income and non-Anglophone settings regarding evidence synthesis, guideline development, and engaged research approaches. Our interviews with interest-holders suggested that when there is low capacity or resources for engagement, guideline developers can also consider engagement during guideline adaptation, seeking input outside of the guideline development group, and consulting literature about population values and preferences.

#### **6.4 Cross-cutting principles and strategies for engagement**

While the findings of the articles included in this thesis are specific to evidence synthesis and health guideline development, many principles for engagement are applicable to health research and decision-making more broadly. Particularly, sharing of power has been identified as a

persistent challenge to overcome in health systems research (Friel et al., 2021; Pratt, 2021; Egid et al., 2021; Topp et al, 2021), and this principle was emphasized in evidence ecosystems. Traditionally, researchers (principal investigators), providers of healthcare, and health policymakers have held decision-making authority throughout evidence synthesis and health guideline development. Adopting an engaged research approach asks these interest-holders to share these positions of power with other groups who may have legitimate interests in health decision-making, such as patients, members of the public, or product makers. While this may put into question the rigour of evidence synthesis or guideline, responsive (or ‘demand-led’) reviews of the literature or health recommendations remain an important way to increase the use of systematically reviewed evidence in decision-making (Langer et al., 2017). Therefore, the concept of rigour should not only include methodological alignment with established gold standards, but also the relevance to decision-making contexts and the perceived legitimacy of the evidence synthesis or guideline by the intended end-user (Langer et al., 2017). However, merely recognizing power differences between interest-holders does not tell us how to navigate them appropriately. Introducing strategies to promote power-sharing between interest-holders, such as including interest-holders in governance and throughout key steps of the project, can help promote equitable engagement of interest-holders and ensure that their perspectives are reflected in final health-related decisions.

Additional challenges also remain in terms of how to assign legitimacy to the evidence provided by interest-holders (Cairney & Oliver, 2017). Indeed, there are paradigmatic tensions between evidence-based medicine, which traditionally places more weight on empirical quantitative evidence, and participatory research approaches. The articles included in this thesis identified persistent issues regarding the devaluation of patient experiential knowledge and a need for co-production in evidence ecosystems. Co-production democratizes the research process, and its principles demand that interest-holders work together as equal partners through shared decision-making, mutual respect, and the integration of diverse perspectives and expertise to produce more relevant and equitable health research (Hickey et al., 2018). Patient and public experiences are often perceived as “less than” empirical evidence, and our interviews with interest-holders suggested that a shift towards epistemic justice is needed within evidence-based medicine which promotes fairness and equality in the production, dissemination, and validation of knowledge.

However, Jull and colleagues (2019) suggested that a focus on equal engagement of interest-holders may be less important than a focus on equitable engagement and the quality of engagement in research processes. Evidence ecosystems should be seen as a site of negotiation between different interest-holders, which include the scientific evidence base and the perspectives of patients, providers of care and other interest-holders. Successful engagement therefore requires pragmatism, combining scientific evidence with principles of governance and co-production to translate complex evidence into narratives that inform health decision-making (Cairney & Oliver, 2017).

To support engagement, guideline developers and evidence synthesis researchers will need to make pragmatic decisions about the resources and boundaries available for co-production in any given project. Engagement is resource-intensive, and providing technical and practical support will be necessary for it to be meaningful and successful. Interest-holders may have different goals and capacity for engagement, and these preferences should be taken into consideration. Examples of support to promote engagement include offering financial compensation, providing training on assessing and summarizing evidence, and having a point-person who can guide interest-holders through technical and complex processes. Having a chair, moderator, or facilitator who can mitigate group dynamics and power imbalances in discussions is essential. This suggests that engagement in research or guideline development does not necessarily happen organically - it requires investments into developing particular responsibilities, skills and attributes to broker the knowledge and implementation processes (Cranley et al., 2017; Flenady et al., 2022). While some evidence is available on facilitator attributes theorized to be associated with successful knowledge translation in health contexts (Elledge et al., 2018), more research is needed on applying these attributes to evidence synthesis and guideline development. Presently, INGUIDE (<https://inguide.org/>) is the first professional certification program in guideline development that offers training for individuals participating in a guideline development group in the role of chair.

## **6.5 Implications for population health**

### **6.5.1 Practice**

The findings of this thesis have widespread applicability to any organization or government that produces evidence syntheses, health recommendations or guidelines, including health technology assessments. We expect the findings to be considered by international organizations such as the World Health Organisation (WHO), Cochrane, Joanna Briggs Institute (JBI), Grading of Recommendations, Assessment, Development, and Evaluations (GRADE), and the Guidelines International Network (GIN). Several knowledge translation activities are planned by the MuSE Consortium to promote uptake of the findings, including publications, presentations and workshops.

Engagement of interest-holders in evidence ecosystems can help ensure that health decision-making is informed by evidence that is relevant, useful and considers diverse perspectives. However, engagement requires an important shift in power structures and governance. Guideline developers and evidence synthesis researchers should do their best to promote equitable engagement by engaging interest-holders as early as possible in the process, providing them with rightful acknowledgment, being clear about expectations and timelines, and facilitating their engagement throughout the process. Organizations may need to make pragmatic decisions as to who they engage, considering their diversity across PROGRESS characteristics and their representativeness, how they are engaged, including at which stage of the process, and with what decision-making power.

Across all articles, very little information was available from equity-deserving groups and low-income settings. It remains unclear whether there are particular conditions or settings (e.g., public health vs health system; high-income vs low income) that influence the engagement of diverse interest-holders. Constraints may exist when rapid guidance is needed or in settings with poor capacity for evidence synthesis and guideline development. To decolonise evidence ecosystems, we should encourage the use of alternatives to the term “stakeholder”, and the MuSE Consortium has proposed “interest-holder” as a suitable replacement (Akl et al., in

progress). Further, international organizations should consider engaging representatives from global south and global north countries and offer capacity building training when necessary. Guideline and evidence synthesis producers should consider the use of checklists and tools, such as the upcoming MuSE extension of the GIN-McMaster GDC for engagement (Petkovic et al., submitted; Magwood et al., in progress; Petkovic et al., 2020) to implement equity promoting practices in synthesis and guideline development processes.

### **6.5.2 Future research**

Based on the findings of this thesis, several areas for future research may be considered. Considering the broad perspective of evidence ecosystems, it is important to evaluate how interest-holder engagement in evidence synthesis and health guideline development influences evidence uptake into health decision-making. Long-term research is necessary to determine if these practices will truly influence health inequities.

Representation and representativeness remain critical issues to address. Particularly, how should “representative” be defined and what are the necessary skills or experiences required for an individual to represent an interest-holder group? Further investigation into the strategies and opportunities to engage people with diverse PROGRESS characteristics is needed, including how to engage people from diverse socioeconomic positions, language abilities, and levels of education. Capacity building opportunities should also be identified for engaging interest-holders from low- and middle-income countries and non-Anglophone countries.

Additionally, much remains to be learned about how to manage diverging disciplinary perspectives, how to recruit stakeholders with diverse characteristics and experiences, or how to sustain stakeholder engagement to support future updates. Future research should explore the perspectives of interest-holders from an array of settings and research/policy domains to determine the most effective practices to support their engagement. The majority of the current evidence base comes from the perspectives of providers of care, patients, members of the public and principal investigators. Additional investigation on the perspectives of program managers,

policymakers, and payers of health research and health services would enhance the current findings.

Finally, future guidance should be developed for alternative approaches to engagement when full co-production and power-sharing is not possible. Many evidence synthesis and guideline development teams face financial and human resource constraints which limit their ability to extensively engage interest-holders. Engaging interest-holders in adaptation processes was identified by our research as one possibility for ensuring that their perspectives are considered in low-resource settings, however more extensive development of these methods is warranted. In our interviews with interest-holders, conducting evidence syntheses on patient values and preferences or conducting surveys was identified as alternative opportunities to promote the patient-centeredness of guidelines. Our qualitative evidence synthesis also recognized existing tools which could promote equitable engagement, such as the use of Delphi processes (Armstrong et al., 2019; Grant et al., 2021; Khodyakov et al., 2019) or the FACE approach for engagement in the development of guideline implementation considerations (Pottie et al., 2021). Given the significant barriers to engagement identified by our qualitative evidence synthesis, these alternative approaches should be acknowledged as they may play an important bridging role toward full co-production of guidelines.

## **6.6 Conclusions**

The engagement of interest-holders in health research and decision-making is growing in popularity and is becoming an expectation in many health systems around the world. Interest-holder engagement helps to bridge the gap between research and practice, ensuring that health policies and programs are better tailored to real-world contexts and the needs of the populations they aim to serve. Focusing on equitable engagement is essential because it ensures that all voices, especially those from marginalized and underrepresented communities, are heard and valued in decision-making processes. This inclusivity helps address power imbalances and social inequities, leading to more just and effective health interventions and outcomes. When engagement is equitable, it promotes trust, enhances the legitimacy of decisions, and promotes

the fair distribution of benefits. While many barriers to engagement exist, these can be meaningfully addressed through the adoption of principles and associated strategies to facilitate the engagement of a broad array of interest-holders. We encourage evidence synthesis researchers and guideline developers to apply these principles, evaluate their strategies, and report on their engagement approaches to further enhance these findings.

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