Patient engagement for the development of equity-focused health technology assessment (HTA) recommendations in the digital era

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Abstract

Background: Health technology assessment (HTA) is a form of policy analysis to inform recommendations for decision-makers. An equity-focused HTA recommendation consists of one that explicitly addresses the impact of health technologies on individuals disadvantaged in society because of their social conditions. However, there is a need for more evidence on the relationships between patient engagement and the development of equity-focused HTA recommendations.

Objectives: The objectives of this dissertation were to examine the association between patient engagement and equity-focused HTA recommendations and identify implementation considerations for patient engagement in HTA.

Methods: I used explanatory sequential mixed methods to analyze 60 HTA reports and 11 interviews with patients and analysts from Canadian organizations: the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO).

Results: Quantitative analysis of the HTA reports showed that patient engagement significantly predicts equity-focused HTA recommendations (OR: 0.26; 95% CI: [0.16 – 0.41]). HTA reviews where HTA analysts directly interviewed patients (OR: 3.85; 95% CI: [2.40 – 6.20]) and where an advisory committee used consensus were more likely to contain equity-focused recommendations (OR: 2.27; 95% CI: [1.35 – 3.84]). Qualitative analysis of the interviews identified strategies for engaging diverse patients in HTA.

Conclusion: The findings of this dissertation can inform the designing of patient engagement in HTA.

Executive summary

Health technology assessment (HTA) represents a form of policy analysis that informs policy-makers decisions about funding and scaling up health technologies to improve health outcomes (O'Rourke al., 2020; Stevens et al., 2003). Organizations, such as the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO), develop HTA recommendations by examining available evidence on effectiveness, cost-effectiveness and safety (O'Rourke al., 2020; Stevens et al., 2003). An equity-focused HTA recommendation consists of one that explicitly addresses the impact of health technologies on individuals disadvantaged in society because of specific health needs or their social conditions (Benkhalti et al., 2021; Braveman & Gruskin, 2003; Tugwell et al., 2006a). HTA recommendations inform policies such as drug coverage, healthcare services, preventive interventions, and public health workforce training, all of which have health equity implications when rolled out to the public.

Studies reported patient engagement as a practical approach for incorporating patient perspectives in recommendations to assist decision-makers in their appraisals of health technologies (Bellemare et al., 2018; O'Flaherty et al., 2021). Patient engagement involves partnerships between HTA institutions and individual patients or patient groups (Facey Helle et al., 2017; Manafo et al., 2018). However, there is a need for more evidence on the relationships between patient engagement processes and the development of equity-focused HTA recommendations.

With the assumption that HTA recommendations serve as a formal knowledge base for stakeholders involved in implementing health technologies, they examine approaches to enhance the integration of health equity factors in those recommendations (Allen, Liberti, et al., 2017).

Furthermore, patients are demanding more accountability from health authorities and calling for considering their voices in decisions regarding their health (Biddle et al., 2021; de Wit et al., 2020; Staley, 2015). Digital technologies offer opportunities for implementing inclusive patient engagement strategies to enhance the development of equity-focused HTA recommendations (Cordoş et al., 2017; Jenssen et al., 2016).

This thesis examines the association between patient engagement processes and equity-focused HTA recommendations and identifies implementation considerations for patient engagement in HTA. I address the objectives by answering the following research questions:

- What are the characteristics of patient engagement processes in HTA?
- What are the characteristics of equity-focused HTA recommendations?
- What patient engagement processes are associated with equity-focused HTA recommendations?
- How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?
- How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?

I used an explanatory mixed methods approach to answer the above questions. For the quantitative portion of the thesis, I analyzed 60 HTA reports from CADTH and HQO. For the qualitative part, I interviewed 11 patients, and HTA analysts involved in patient engagement at CADTH and HQO. I obtained ethics approval from the University of Ottawa to conduct the qualitative study.

Overview of findings

Characteristics of patient engagement processes: For HQO reports, HTA analysts interviewed patients to obtain their input. For CADTH reports, patient organizations responded to calls for input that CADTH posted on their website. HTA and patient organizations used various methods to engage patients, including interviews, focus groups and surveys. The primary modality of engagement consisted of digital technologies. However, HTA organizations and patient groups alike held in-person meetings. Patients contributed to decision-making to formulate HTA recommendations as members of HTA advisory committees. Decision-making models varied depending on the types of HTA reviews. CADTH used voting for common drugs, whereas they used consensus for oncology drugs. HQO used consensus in decision-making to develop HTA recommendations.

Characteristics of equity-focused HTA recommendations: An equity-focused HTA recommendation consisted of a recommendation that contained at least one PROGRESS-Plus item. PROGRESS-Plus stands for Place of residence, Race/ ethnicity, Occupation, Gender/sex, Socioeconomic status, and Social capital. The Plus designates other strata, such as sexual orientation and individuals with disabilities, used to describe participants in health-related research and interventions. Most reports had equity-focused HTA recommendations (68%; 41 out of 60). There were 12 unique PROGRESS-Plus items across all the included HTA reports. Six items were from the PROGRESS category: the place of residence, language, gender, education, socioeconomic status, and social capital. The other six items were from the "Plus" category: affordability, age, ethical issues, the severity of the conditions, logistics of treatment, and stigma. There was no record of the following items from the PROGRESS framework: occupation, race/ethnicity/culture, and religion.

Association between patient engagement processes and equity-focused HTA recommendations: Quantitative analysis of the HTA reports showed that direct patient engagement (OR: 3.85; 95% CI: [2.40 – 6.20]) and consensus in decision-making (OR: 2.27; 95% CI: [1.35 – 3.84]) were more likely to be associated with the development of equity-focused HTA recommendations in comparison to indirect patient engagement (OR: 0.26; 95% CI: [0.16 – 0.41]) and voting (OR: 0.44; 95% CI: [0.26 – 0.73]). Qualitative analysis of the interviews identified strategies for engaging diverse patients in HTA.

Qualitative analysis of interviews with five patients: The interviewees said that HTA organizations should provide practical support to patients and HTA practitioners, including travel accommodations, compensation, and training. The interviewees also advised HTA organizations to use digital alongside non-digital tools to support inclusive engagement strategies. The interviewees indicated that HTA organizations should consider the digital divide, which may arise from patients' social environment and preferences.

Qualitative analysis of interviews with six HTA analysts: The interviewees shared about current practices that allowed them to successfully reach out, recruit and engage underrepresented groups in their HTA. The interviewees also highlighted the need to raise awareness about the importance of patient input alongside clinical evidence to inform health equity analysis in HTA. The interviewees discussed the role of digital technologies in strengthening organizational readiness for inclusive and diverse patient engagement. However, the interviewees cautioned that HTA organizations should consider the digital divide in broader strategies to improve patient engagement in HTA.

Conclusion

To my knowledge, the dissertation is the first study that quantifies the relationship between patient engagement and the development of equity-focused HTA recommendations. The study also reveals that contextual and procedural factors, such as availability of data, awareness about patients' role in HTA, and use of digital technologies, may impact this relationship. As in previous studies (Black et al., 2018; Roche et al., 2020), the findings reiterate patient engagement's importance in identifying health equity factors in HTA. The findings of this dissertation can inform strategies for advancing health equity through HTA with patient engagement. The results have implications for practice, research, policy, and education.

Contribution of the authors

The Ph.D. candidate, Rosiane Siméon, prepared three manuscripts as part of this dissertation (Chapters 5-7). I will submit all the manuscripts for publication. Rosiane Siméon is the first author of all the manuscripts; the supervisors and advisory committee members are all co-authors. Another author, a Ph.D. candidate, contributed to the qualitative analysis. Additional authors may be confirmed later before the publication of the quantitative study. They are students who helped collect data for the quantitative research. Below, I describe the contribution of the confirmed co-authors:

- Rosiane Siméon (RS): is the Ph.D. candidate and led the research planning, study design, data collection, analysis, and writing of the manuscripts.
- Drs. Peter Tugwell and Vivian A. Welch: co-supervised the Ph.D. candidate and contributed to the research planning, study design, and review of the manuscripts.
- Drs. Ian D. Graham, Kumanan Wilson, Reiner Banken, Shehzad Ali, and Professor Janet Hatcher Roberts are the Thesis Advisory Committee (TAC) members and contributed to the study design and review of the manuscripts.
- Ms. Anita Rizvi (AR): is a fellow Ph.D. candidate and contributed to the qualitative analysis in Chapters 6 and 7.

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Dedication

To my parents -Zilitha and Romusset, and my son -Nolann, for inspiring me.

Table of Contents

List of Tables

List of Figures

Chapter 1: Introduction

1.1 Overview of Chapter 1

In this introductory chapter, I define key concepts and discuss background information for the dissertation. I also briefly describe my methodology with a summary table about the research design to answer each question. I conclude the chapter with an overview of the reporting structure of the dissertation. As in many articles reviewed for this dissertation, I use the terms patient engagement and patient involvement interchangeably.

1.2 Background

Decision-makers can achieve health equity by acting on modifiable factors that cause unfair distribution of health outcomes across populations (Braveman & Gruskin, 2003; Marmot, 2017). Researchers propose various tools to facilitate such modifications to pursue health equity, including knowledge production (Tugwell et al., 2006a), practice guidelines (V. A. Welch et al., 2017a), intervention development (O'Neill et al., 2014), and policy analysis (Benkhalti et al., 2021). Health technology assessment (HTA) represents a form of policy analysis that informs policy-makers decisions about funding and scaling up health technologies to improve health outcomes. Organizations, such as the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO), develop HTA recommendations by examining available evidence on the effectiveness, cost-effectiveness and safety of health technologies (O'Rourke al., 2020; Stevens et al., 2003).

Health technologies are inherent in health service infrastructure and include pharmaceuticals and non-pharmaceutical products such as diagnostic, healthcare services, preventive, treatment and rehabilitation procedures to support health and well-being (O'Rourke et al., 2020; Stevens et al., 2003). Regulatory agencies such as Health Canada and the Food and

Drug Administration (FDA) authorize health technologies (Gutiérrez-Ibarluzea et al., 2017; Menon, 2015). However, at this stage, the health technology may be only accessible to a relatively small group of patients, while it may have the potential to benefit a larger pool of individuals (Angelis et al., 2018; Gutiérrez-Ibarluzea et al., 2017). HTA is then conducted during this post-market phase to inform decisions about increasing its accessibility through funding by health insurance and other drug accessibility programs (Angelis et al., 2018; Gutiérrez-Ibarluzea et al., 2017; O'Rourke., 2020).

Initially, researchers proposed HTA as a guide for expert decisions on ensuring that health technologies are safe for use and meet efficiency goals (Allen, Liberti et al., 2017; Stevens et al., 2003; Sullivan et al., 2009). Lately, HTA organizations have highlighted the importance of health equity in their vision (Busse et al., 2015; Guindo et al., 2012; Hill & Olson, 2014). This interest in health equity is occurring as health systems are moving from an expert-controlled environment to a more patient-driven culture (Edgman-Levitan & Schoenbaum, 2021; Swan, 2009). Growing awareness about patient rights and easy access to information through digital technologies also power this culture of patients voicing their concerns and sharing their experiences with health technologies (Kent & Yellowlees, 2015; Mazanderani et al., 2013). With the assumption that HTA recommendations serve as a formal knowledge base for stakeholders involved in implementing health technologies, examining approaches to enhance the integration of health equity considerations in those recommendations is essential (Allen, Walker, et al., 2017).

An equity-focused HTA recommendation consists of one that explicitly addresses the impact of health technologies on individuals disadvantaged in society because of specific health needs or social characteristics (Benkhalti et al., 2021; Culyer & Bombard, 2012). As such, an equity-focused recommendation puts those with higher risks of experiencing adverse health outcomes at the center of the HTA issue. Studies identified the disparate and inequitable distribution of health outcomes based on socially constructed characteristics such as those described in the PROGRESS-Plus framework (O'Neill et al., 2014). PROGRESS-Plus stands for Place of residence, Race/ ethnicity, Occupation, Gender/sex, Socioeconomic status, and Social capital. The Plus designates other strata, such as sexual orientation and individuals with disabilities, used to describe participants in health-related research and interventions.

Patient engagement involves a partnership between researchers and patients to capture patient perspectives and help patients influence knowledge creation, like HTA recommendations (Facey Helle et al., 2017; Frank et al., 2020; Higgins et al., 2017). HTA organizations increasingly adopt patient engagement as a practical approach for incorporating patient perspectives in developing recommendations to assist decision-makers in their appraisal of health technologies (Bellemare et al., 2018; O'Flaherty et al., 2021). Some HTA organizations have made patient engagement mandatory in their HTA process (Haerry et al., 2018). In practice, HTA analysts engage individual patients or patient groups to inform their work (Facey Helle et al., 2017; Manafo et al., 2018; Wahlster et al., 2017a). In HTA research, a patient consists of an individual with one of the following attributes: living with a health condition, caregivers of individuals living with a health condition, advocate who supports people living with a specific health condition, expert who has experience with and technical knowledge about a health condition (Facey Helle et al., 2017; Higgins et al., 2017; McCoy et al., 2019).

Patient groups are generally member-based organizations composed of individuals who share everyday experiences with a health condition or a disease (Hicks et al., 2014). The primary purpose of patient groups is to ensure that patient perspectives are reflected in decisions regarding their health (Hicks et al., 2014; Maguire & Britten, 2017). In the context of HTA, their work covers various interests, including effectiveness, legal and ethical issues, accessibility, and pricing of health technologies (Facey Helle et al., 2017; McCoy et al., 2019). Patient groups can be locally grown or as branches of bigger international and national patient advocacy groups (Facey Helle et al., 2017). It is important to note that advocacy approaches between patients and public interest groups may differ in HTA. Public interest groups usually have a similar purpose as patient groups, yet they tend to focus more on finding balance in using public money to fund health technologies (Facey Helle et al., 2017; McCoy et al., 2019; Moreira, 2015). Other public interest groups may be more preoccupied with representing societal values in HTA recommendations (Facey Helle et al., 2017; Moreira, 2015).

Digital technologies cover various communication tools for data production, management, and processing (Aziz & Madani, 2015; Mitchell & Kan, 2019). Digital technologies can enable social interactions, facilitate access to resources and asynchronously enhance the sharing of information using various levels of interactions (Aziz & Madani, 2015; Mitchell & Kan, 2019). In population and public health, digital technologies help deliver services to the public, connect community members, support professional training and promote public health (Jackson et al., 2018; Petkovic et al., 2021; Wilson & Flood, 2021).

1.3 Statement of the problem

In the context of HTA, studies reported that patient engagement is essential for capturing lived experiences with health technologies (Douglas et al., 2015; Facey Helle et al., 2017). Several studies also highlighted the role of patient engagement in understanding the effectiveness of health technologies (Bellemare et al., 2018; Douglas et al., 2015; Facey et al., 2018; Higgins et al., 2017; Staley, 2015). Research shows that differences in perceptions about the meaning of health and experiences with society can shape the framing of health problems among population groups (Brüssow, 2013; Charlier et al., 2017; Staley & Doherty, 2016).

As for most technologies, health technologies may not necessarily reflect the needs and experiences of many population groups (AlQudah et al., 2021; Hill & Olson, 2014). Also, clinical trials may not provide sufficient evidence about the effect of health technologies because some patient groups are underrepresented in research for various reasons (Armstrong et al., 2017; Forsythe et al., 2019; Tugwell et al., 2006a). Certain individuals are underrepresented in biomedical research because they have been historically disadvantaged in society (Ellard-Gray et al., 2015). Studies also reported research design, ethics, and funding as barriers to including specific population groups in health research (Hanney et al., 2004; Jacklin & Kinoshameg, 2008; Shivayogi, 2013). These populations may consist of individuals with disabilities, people living in remote areas, and individuals treated for multiple illnesses (Frohlich & Potvin, 2008; Jacklin & Kinoshameg, 2008). The underrepresentation of certain groups implies that researchers cannot establish the efficacy of health technologies in those individuals.

The above examples have health equity implications because they depict challenges that can delay or prevent patients from accessing health technologies to improve health outcomes. Institutions envisioning health technology assessment (HTA) as a process leading to health equity must consistently adapt their patient engagement strategies. Equitable patient engagement processes will allow HTA organizations to face challenges that arise from growing recognition of democratic rights (Safaei, 2015a), easy access to information (Kingod et al., 2016), population mobility (van der Waal et al., 2017), and the shift to online interactions (Khalili et al., 2020).

1.4 Research gaps

As shown in Figure 1, I conducted a narrative review that revealed several research gaps around patient engagement to develop equity-focused HTA recommendations. Chapter 2 is a full report of the narrative review. I only discuss the research gaps in the thesis in the following paragraphs.

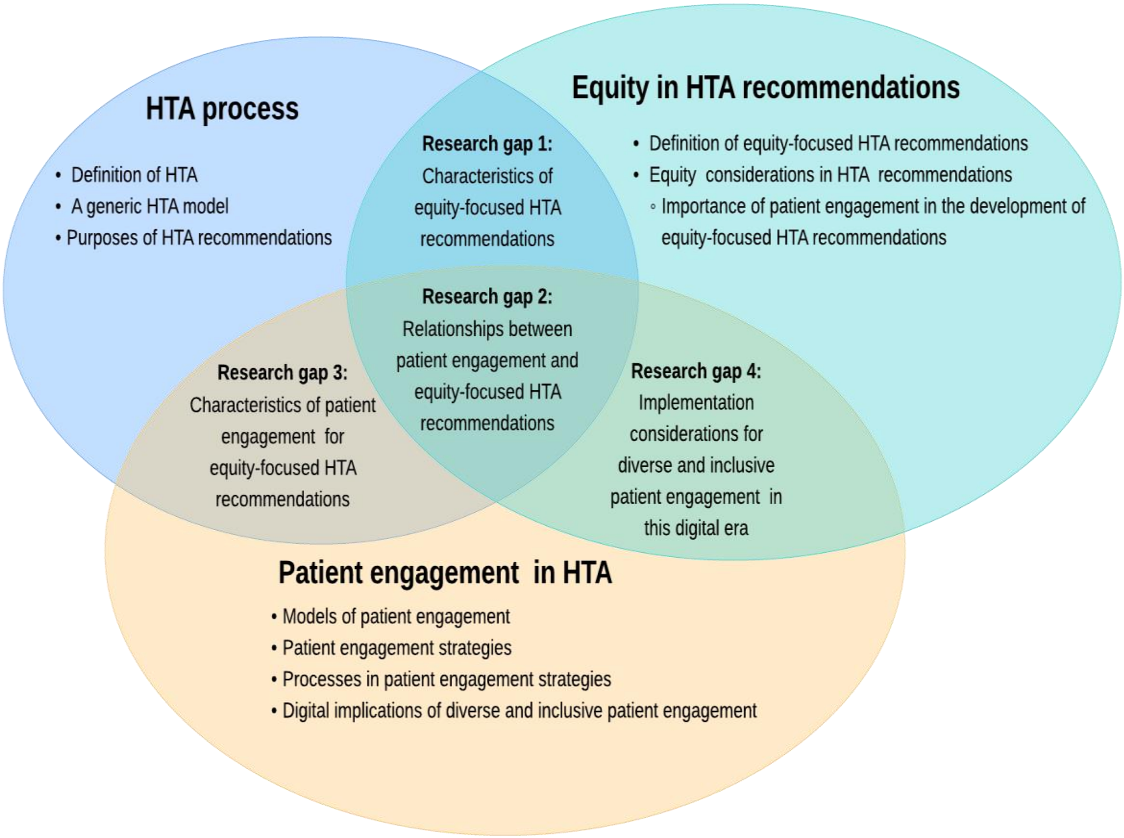

Figure 1: Research gaps from the literature

1.4.1 Research gap 1: Characteristics of equity-focused HTA recommendations

Characterizing health equity factors in HTA recommendations can help draw attention to the need to systematically conduct health equity analysis in HTA (Benkhalti et al., 2021; Hosking et al., 2019; Prady et al., 2018). Researchers developed tools to help incorporate and report health equity considerations in HTA (Benkhalti et al., 2021; Culyer & Bombard, 2012; Hoch et al., 2021). For example, Benkhalti et al. 2021propos2021) proposed a checklist for health technology assessment (ECHTA) that helped HTA analysts identify risks of health inequities across various population groups impacted by a health technology implemented in Quebec, Canada. Other researchers have attempted to develop search filters for identifying equity-focused studies (Hosking et al., 2019; Prady et al., 2018).

Combining specific terms to label social determinants of health and non-specific terms to differentiate the socio-demographic characteristics of research participants can help identify equity-focused studies more accurately. In addition to parameters described in the PROGRESS-Plus framework for characterizing population groups, some researchers recommend supplementing these factors with non-specific terms referring to group comparison, such as: "availability or lack of alternative options," "risk factors for" "were high/low" "at higher risk for" "significant among" and "differed by" (Benkhalti et al., 2021; Hosking et al., 2019; Prady et al., 2018).

1.4.2. Research gap 2: Relationships between patient engagement and equity-focused HTA recommendations

Evidence is growing on the importance of patient engagement in HTA (Bidonde et al., 2021; Facey et al., 2018; Gunn et al., 2021; Scott et al., 2017). However, there is a need for more research that examines associations between patient engagement processes and the incorporation of equity considerations in HTA recommendations (de Wit et al., 2020; Facey, 2019; Mercer et al., 2020). Similarly, there is a need to improve existing engagement processes to increase the contribution of patients with diverse backgrounds in HTA (Majid & Gagliardi, 2019; Scott et al., 2017; J. L. Wale et al., 2021). The research gaps mentioned above can further increase uncertainty in developing HTA recommendations. Thus, research that can help address these gaps can contribute to advancing the fields of HTA and population health.

It is essential to acknowledge that multiple strategies can help incorporate equity factors in HTA recommendations (Benkhalti et al., 2021; Busse et al., 2015; Marsh & Le, 2015). For example, the significance and the weight of strategies regarding equity considerations in HTA recommendations will vary depending on governance systems (Espinoza & Cabieses, 2014; Hill & Olson, 2014; Mohara et al., 2012). Patient engagement is only one approach to developing equity-focused HTA recommendations (Cookson et al., 2017; Facey Helle et al., 2017; Snow et al., 2018). Clarifying the relationships between patient engagement and the development of HTA recommendations can reinforce the validity of guidance on implementing health technologies (Dipankui et al., 2015; Staley & Doherty, 2016; J. Wale et al., 2017).

1.4.3. Research gap 3: Characteristics of patient engagement to inform equity-focused HTA recommendations.

Studies have not identified a uniform taxonomy to describe patient engagement methods (Bombard et al., 2018a; Chudyk et al., 2022; Dipankui et al., 2015; Greenhalgh et al., 2019). Strategies for engaging patients in research vary widely with the stage and context of the research (Bombard et al., 2018a; Concannon et al., 2012; Facey Helle et al., 2017; Frank et al., 2020). Patient engagement strategies vary with the context of HTA organizations and the characteristics of the health technology (Facey Helle et al., 2017; Manafo et al., 2018). Patient engagement processes in research and HTA are increasingly gaining attention (Biddle et al., 2021; Chudyk et al., 2022; Davis et al., 2019; de Wit et al., 2020; Facey, 2019; Greenhalgh et al., 2019). A recent scoping review identified 14 models and frameworks of patient engagement in health-related research (Chudyk et al., 2022). The study identified 18 overlapping and 57 divergent elements across six conceptual categories (Chudyk et al., 2022). Considering this abstract landscape, it becomes essential to determine which procedural features best support the purpose of patient engagement (Bidonde et al., 2021; Chudyk et al., 2022; de Wit et al., 2020; Frank et al., 2020; Greenhalgh et al., 2019).

1.4.4. Research gap 4: Implementation considerations for diverse and inclusive patient engagement

Researchers described diversity in patient engagement as "bringing less traditional voices" to health research (Davis et al., 2019; Dawson et al., 2018; Khuntia et al., 2022; Roche et al., 2020). Less traditional voices may include patients facing barriers to research participation because of their living conditions, geographical locations, and physical and mental health conditions (Gill et al., 2018; Reynolds et al., 2021; Roche et al., 2020). However, achieving diversity in the representation of population groups comes with several challenges, including identifying patient representatives and accommodating patients (Domecq et al., 2014; Dukhanin et al., 2020; Maguire & Britten, 2017). Some researchers advised focusing on collecting diverse perspectives to compensate for the lack of patient representation in studies ((Crockett et al., 2019; de Wit et al., 2020; Higgins et al., 2017; Maguire & Britten, 2017; Reynolds et al., 2021; Shimmin et al., 2017; Snow et al., 2018).

Researchers also called for applying health equity and social justice lens in patient engagement strategies to help increase diverse perspectives in health research (Davis et al., 2019; Dawson et al., 2018; Roche et al., 2020; Shimmin et al., 2017). Studies showed that inclusive engagement could elicit diverse perspectives to impact research narratives (Dawson et al., 2018; De Santis et al., 2019; Reynolds et al., 2021; Roche et al., 2020). Researchers described inclusive patient engagement as creating a research environment where research teams manage power dynamics and research processes to enable patients to contribute positively and productively to the research activities (Black et al., 2018; Reynolds et al., 2021; Shimmin et al., 2017). There is a need to identify strategies to increase the diversity of perspectives to inform the development of equity-focused recommendations (Black et al., 2018; de Wit et al., 2020).

1.4.5. Digital implications of implementing diverse and inclusive patient engagement

Digital technologies help to enable broader citizen engagement and bring diverse perspectives into mainstream debates about health technologies (Kent & Yellowlees, 2015; Mazanderani et al., 2013; Mitchell & Kan, 2019). HTA organizations frequently use digital technologies such as websites, webinars, and social media to support stakeholder engagement in their processes (Facey Helle et al., 2017; Jenssen et al., 2016; Liddy et al., 2017). As digital technologies continually transform how patients interact with their environment, it becomes crucial to explore how its potential can leverage the contribution of patients from diverse backgrounds in the HTA process (Doupi, 2016; Kent & Yellowlees, 2015; Mitchell & Kan, 2019).

Furthermore, patients are demanding more accountability from health authorities and calling for considering their voices in decisions regarding their health (Biddle et al., 2021; Crockett et al., 2019; de Wit et al., 2020; Snow et al., 2018; Staley, 2015). Digital technologies offer opportunities for implementing diverse and inclusive patient engagement strategies, which can enhance the development of equity-focused HTA recommendations (Cordoş et al., 2017; Jenssen et al., 2016; Mitchell & Kan, 2019). HTA recommendations inform policies such as drug coverage, healthcare services, preventive interventions, and public health workforce training, all of which have health equity implications when rolled out to the public.

1.5 Research objectives

The dissertation examines the relationships between patient engagement processes and equity-focused HTA recommendations and identifies implementation considerations for patient engagement in HTA. The research questions are:

1. What are the characteristics of patient engagement processes in HTA?
2. What are the characteristics of equity-focused HTA recommendations (frequency and types of PROGRESS-Plus items recorded in HTA recommendations)?
3. What patient engagement processes are associated with equity-focused HTA recommendations?
4. How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?
5. How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?

1.6 Methodology

1.6.1. Research design: explanatory sequential mixed methods

I used an explanatory sequential mixed methods approach to answer the above research questions. This approach uses findings from quantitative data analysis to inform data collection for subsequent qualitative analysis (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017). I selected this research approach because it can reveal relationships between variables while elucidating the context and meanings of the findings with research participants (Regnault et al., 2018; Shorten & Smith, 2017; Vedel et al., 2019). As illustrated in Figure 2, I analyzed each study separately; I used the results in an integrated discussion to address the research objectives (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017). Also, the qualitative and quantitative results have equal value in helping expand current evidence on the topics covered (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017).

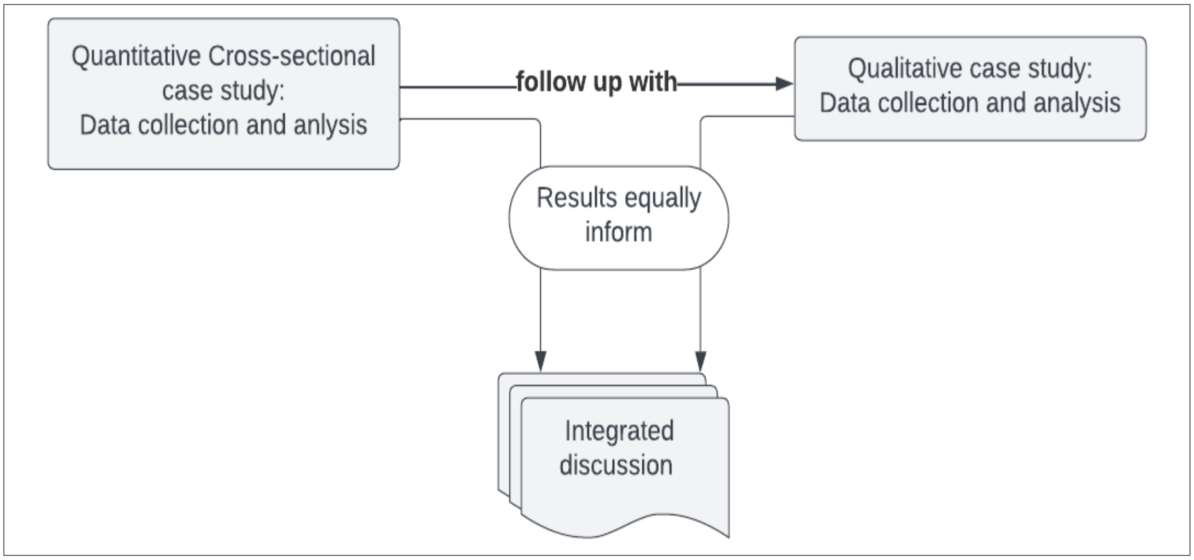

Figure 2: *Explanatory sequential mixed methods design to the dissertation*

1.6.2. Philosophical worldview: pragmatic worldview

I used a pragmatic research approach because it emphasizes applying research to tackle a specific problem (Creswell & Clark, 2017; Kaushik & Walsh, 2019). The pragmatic philosophy of research helps frame the production of knowledge from the perspectives of the research participants(Kaushik & Walsh, 2019). It also acknowledges that truth is context-bound and varies with one's perspective, a constructivist paradigm- encompassing the social construction of reality (Kaushik & Walsh, 2019).

1.6.3. Researcher positionality statement

I chose this research approach because it enables participants to share their perspectives and impact research narratives (Creswell & Clark, 2017; Kaushik & Walsh, 2019; Schoonenboom & Johnson, 2017). It also provides flexibility in using multiple methods for data collection and engaging research participants to help reveal their perspectives (Creswell & Clark, 2017; Kaushik & Walsh, 2019). Moreover, it promotes collaboration between researchers and research participants (Kaushik & Walsh, 2019), facilitating the use of research findings (Graham et al., 2006). Additionally, the pragmatic worldview is well-aligned with the focus of the population health field, which has population groups directly affected by health interventions at the center of research for developing policy alternatives (Carpiano & Daley, 2006; Kaushik & Walsh, 2019). Finally, I believe that using a pragmatic approach in this dissertation also acknowledges the diversity of needs and interests amongst patients, HTA analysts, HTA researchers and decision-makers, who are all critical actors in advancing health equity through their contribution to HTA.

1.6.4. Contribution to the fields of HTA and population health

The field of population health focuses on studying the link between interventions and the health outcomes of a group of individuals (Kindig and Stoddart, 2003). The focus on health equity is essential to the field of population health to address the distribution of health outcomes in individuals living in poorer social conditions (Braveman 2014). Implementing HTA recommendations can affect the distribution of health outcomes based on social characteristics (Stevens et al. 2003).

The thesis contributes to the field of population health by bringing more awareness to how patient engagement processes could influence the development of HTA recommendations to impact the distribution of health outcomes. The dissertation also contributes to advancing the fields of HTA. It highlights the need to systematically evaluate patient engagement processes in HTA to ensure that HTA recommendations consider the concerns of diverse populations. Characterizing patient engagement processes to inform equity-focused HTA can pave the way for developing novel strategies to help advance health equity through HTA involving patients.

1.6.5. Organization of the dissertation

The dissertation is article-based, meaning research results are presented as three manuscripts. In Table 1, I summarize the research questions, methods, data sources and analyses to address the stated research objectives. In terms of reporting structure, the dissertation contains eight chapters. **Chapter 1,** the introduction, contains key definitions of concepts discussed throughout the dissertation. **Chapter 2,** the literature review, consists of a narrative review that discusses current evidence and research gaps around patient engagement to inform health equity concerns in HTA recommendations. **Chapter 3** describes the overall dissertation methods and theoretical approach to guide the research project. **Chapter 4** consists of a brief overview of the manuscripts. **Chapter 5** is the first manuscript of the series and consists of a quantitative study that revealed that certain patient engagement processes could lead to incorporating health equity concerns in HTA recommendations. **Chapter 6,** the second manuscript, consists of a qualitative analysis of patient perspectives on implementation considerations for patient engagement in HTA. **Chapter 7** is the third manuscript that describes additional findings of the qualitative study from the viewpoint of HTA analysts. **Chapter 8,** the integrated discussion, highlights the dissertation's unique contribution of patient engagement to developing equity-focused HTA recommendations. The chapter ends with a conclusion section that relates the implications of the findings for different actors involved in advancing health equity in HTA, including patients, HTA analysts, researchers, and decision-makers. The next chapter covers the literature review.

Table 1. Research objectives, research questions, methods, data sources and analyses

Research objectives (Chapter)	**Research questions**	**Methods and data sources**	**Analyses**
1. Examine relationships between patient engagement processes and equity-focused HTA recommendations (Chapter 5)	1. What are the characteristics of patient engagement processes in HTA? 2. What are the characteristics of equity-focused HTA recommendations? 3. What patient engagement processes are associated with equity-focused HTA recommendations?	1. Quantitative case study: purposeful sample of 60 HTA reports published between 2013 and 2021 from two Canadian HTA organizations, CADTH and HQO.	1. Descriptive analysis: PROGRESS-Plus framework 2. Inferential analysis: odd ratio
2. Identify implementation considerations for patient engagement in HTA (Chapters 6 and 7)	4. How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations? 5. How could HTA organizations use patients in ways that acknowledge the diversity of people and needs impacted by health technologies?	2. Qualitative case study: interviews with five patients aged 18 years and older who participated in patient engagement at CADTH and HQO over the past five years. 3. Qualitative case study: interviews with six HTA analysts from CADTH and HQO.	3. Grounded theory-

Chapter 2: Review of the literature

2.1. Overview of Chapter 2

Chapter 2 is a narrative review that represents the body of evidence that informs the background of the dissertation. For this review, I use peer-reviewed publications, including individual research reports, systematic and non-systematic reviews about patient engagement, and the incorporation of equity factors in HTA recommendations. Also, note that I use the terms patient engagement and patient involvement interchangeably and repeat information from the review -verbatim or paraphrased- across the dissertation chapters.

As shown in Figure 1, the review covers three main topics: the HTA process, equity in HTA recommendations and patient engagement strategies in HTA. I also discuss the digital implications of inclusive patient engagement in HTA. I conclude the chapter with a summary of the results and their impact on research.

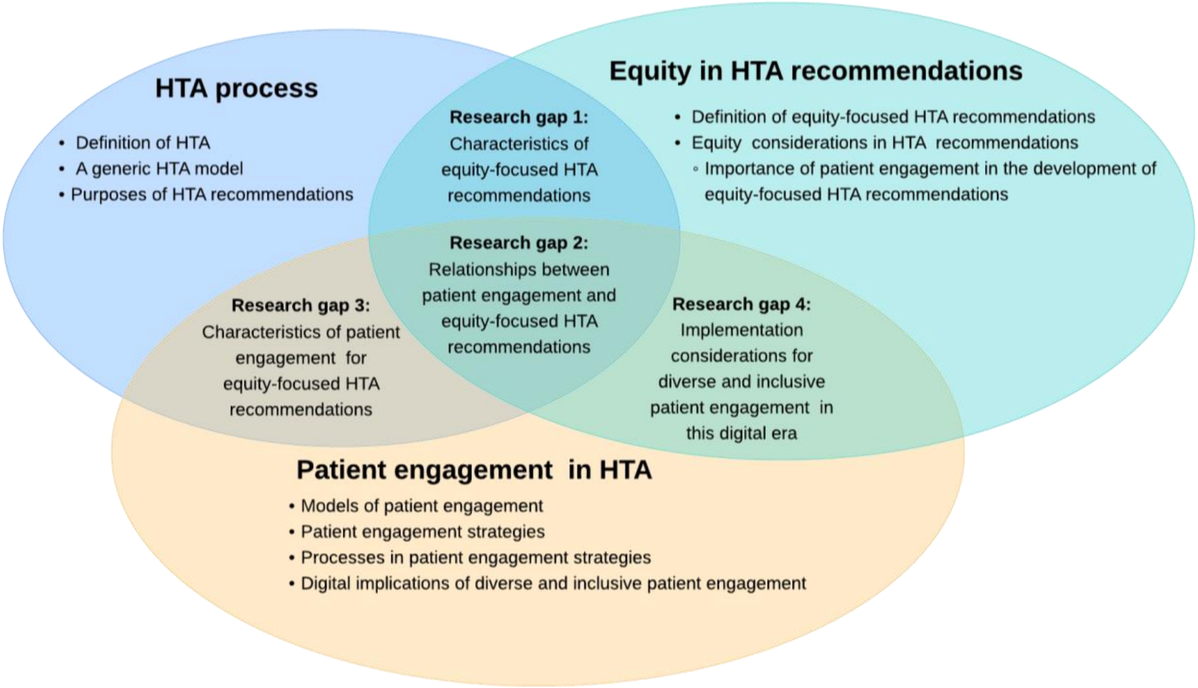

Figure 1: Research gaps from the literature

2.2. Overview of the Health Technology Assessment (HTA) Process

2.2.1. Definition of HTA

Health technology assessment (HTA) represents a form of policy analysis for informing decision-makers about funding and scaling up the implementation of health technologies for improving health outcomes (O'Rourke et al., 2020; Stevens et al., 2003). Health technologies are inherent in health service infrastructure, including pharmaceuticals such as diagnostic, preventive, treatment and rehabilitation procedures used to support health and well-being (O'Rourke et al., 2020; Stevens et al., 2003). In addition, research also described HTA as an iterative knowledge-creation activity where stakeholders such as patients and decision-makers are engaged throughout the process (Wahlster et al., 2017a).

2.2.2. A Generic HTA Model

An integrated approach to HTA comprises five steps: identification of HTA objectives and health technologies, identification of evidence needs, evidence mapping and HTA decision-making. Stakeholder engagement, including patient engagement, is identified as a core element (Wahlster et al., 2017a). As such, HTA organizations can involve patients at either selected or all phases of the HTA process. Figure 3 illustrates a generic model of HTA with critical outputs from each step leading to the development of HTA recommendations (Wahlster et al., 2017a).

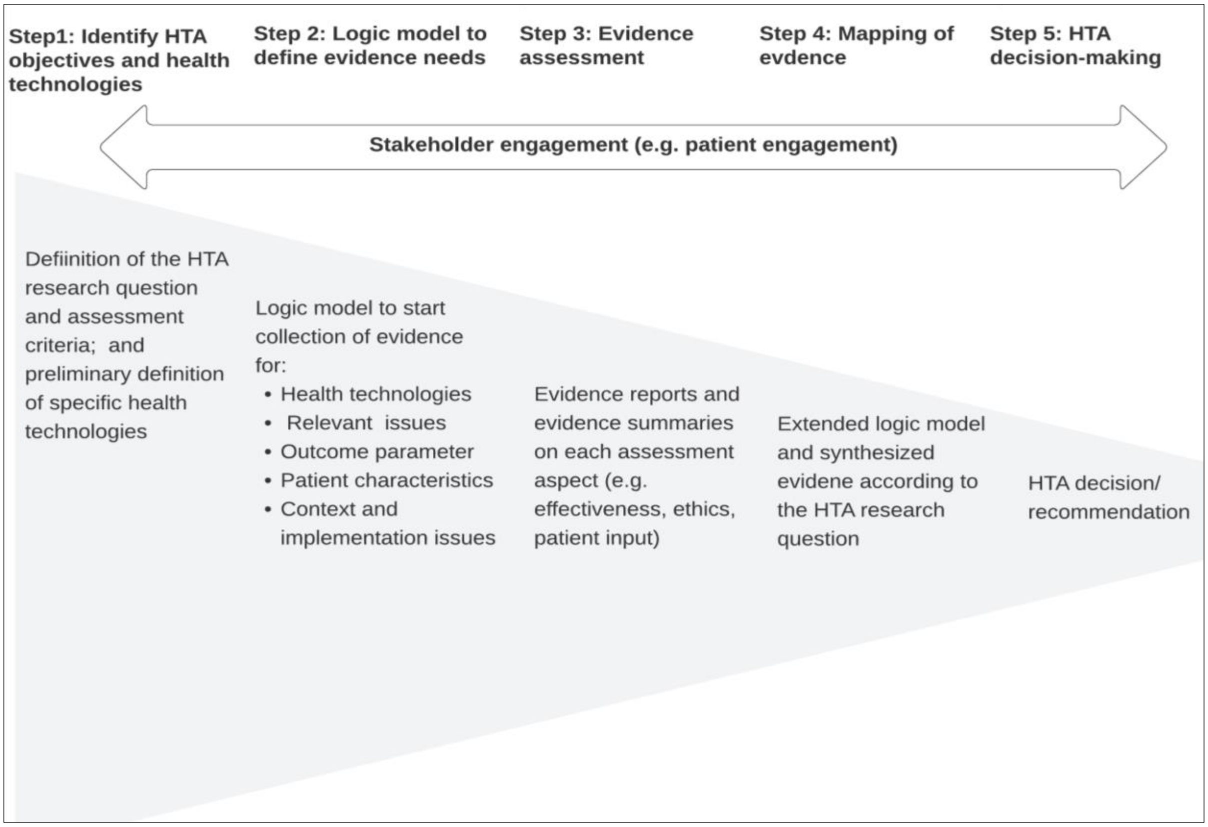

Figure 3. Generic HTA model (adapted from Wahlster et al., 2017)

In general, HTA processes vary depending on available competencies in organizations and the characteristics of healthcare systems (Kõrge et al., 2017; Wahlster et al., 2017). Most HTA organizations follow national guidelines to conduct an assessment and develop recommendations, which may cover one or multiple attributes of health technologies (Kõrge et al., 2017; Pasternack et al., 2009; Wahlster et al., 2017). HTA organizations tend to focus on studying the following attributes of health technologies: effectiveness, cost-effectiveness, quality of life and safety(Pasternack et al., 2009; Wahlster et al., 2017a). Studies also reported that ethical and non-economic criteria also inform decisions in HTA(Bellemare et al., 2018; Wahlster et al., 2017a). HTA recommendations are typically drawn from examining available evidence on health technologies' effectiveness, cost-effectiveness, and safety (O'Flaherty et al., 2021; O'Rourke et al., 2020; Wahlster et al., 2017).

2.2.3. Purpose of HTA recommendations

Studies suggested that HTA recommendations serve various purposes, including conceptual, instrumental, and symbolic use in decision-making regarding the coverage and implementation of health technologies. Conceptual use consists of implementing the HTA recommendations framework for catalyzing change in awareness, enhancing understanding and developing position statements about a specific health condition or technology (Hivon et al., 2005). Instrumental use denotes adopting HTA recommendations to support health technology use, promotion, and adoption (Hivon et al., 2005). Symbolic use refers to using HTA recommendations to justify a position or enforce policies about a specific health technology (Hivon et al., 2005).

Initially, HTA researchers proposed HTA as a guide for expert decisions on ensuring that health technologies are safe for use (Allen et al., 2017; Stevens et al., 2003; Sullivan et al., 200, and meet efficiency goals (Allen, Liberti, et al., 2017; Sullivan et al., 2009). Lately, HTA organizations have highlighted the importance of health equity in their vision (Brown & Wobst, 2021; Busse et al., 2015; Hill & Olson, 2014). This interest in health equity is occurring similarly; health systems are moving from an expert-controlled environment to a more patient-driven health system (Edgman-Levitan & Schoenbaum, 2021; Swan, 2009; J. Wale et al., 2017).

Research gaps: Growing awareness about patient rights and easy access to information through digital technologies also power this culture of patients voicing their concerns and sharing their experiences with health technologies (Kent & Yellowlees, 2015; Mazanderani et al., 2013; Safaei, 2015a). With the assumption that HTA recommendations serve as a formal knowledge base for stakeholders involved in the implementation of health technologies (Hivon et al., 2005, O'Rourke et al., 2020; Stevens et al., 2003), it is appropriate to examine approaches to enhance the integration of health equity considerations in those recommendations.

2.3. Development of equity-focused HTA recommendations

2.3.1. Definition of equity-focused HTA recommendations

I define an equity-focused HTA recommendation as one that explicitly addresses the impact of health technologies on individuals disadvantaged in society because of specific health needs or social characteristics (Benkhalti et al., 2021; Culyer & Bombard, 2012). As such, an equity-focused recommendation puts those with higher risks of experiencing adverse health outcomes at the center of the HTA issue. Aiming to achieve equity in health is generally understood as balancing the benefits and harms of interventions to reduce the burden of adverse health outcomes on socially disadvantaged individuals (Braveman & Gruskin, 2003; Cookson et al., 2017).

2.3.2. Health equity considerations in HTA recommendations

As for most technologies, health technologies may not necessarily reflect the needs and experiences of many population groups (AlQudah et al., 2021; Hill & Olson, 2014). Some individuals are underrepresented in biomedical research because they have been historically disadvantaged in society (Clark & Preto, 2018; Ellard-Gray et al., 2015; Ward et al., 2021). Others may not participate in research for ethical reasons(Clark & Preto, 2018; Macklin, 2010). There are also shortcomings related to critical aspects such as research design, ethics, and funding as fundamental barriers to the representation of specific populations in studies (Facey et al., 2018; Frank et al., 2020; Jacklin & Kinoshameg, 2008; Shivayogi, 2013). These populations may include individuals with disabilities, people living in remote areas, people from low socioeconomic positions, children and individuals being treated for multiple illnesses (Frohlich & Potvin, 2008; Shivayogi, 2013).

Individuals may be disadvantaged compared to others in society because of constraints and stigma associated with sociodemographic characteristics such as race, ethnicity, age, gender, and level of education (Frohlich & Potvin, 2008; Krieger, 2014; Shivayogi, 2013). Some individuals may be disadvantaged because they live in vulnerable circumstances, such as poverty and a degraded environment, which can put them at higher risk of disease than others (Frohlich & Potvin, 2008; Shivayogi, 2013). Other people may be disadvantaged because they live with debilitating illnesses (Frohlich & Potvin, 2008; Shivayogi, 2013).

Disparate and inequitable distribution of health outcomes were identified in studies based upon socially constructed characteristics such as those described in the PROGRESS-Plus framework (O'Neill et al., 2014). PROGRESS stands for Place of residence, Race/ ethnicity, Occupation, Gender, Religion, Education, Socioeconomic status, and Social capital. The Plus designates any other reported strata, sexual orientation, and disabilities. Studies also described barriers associated with these factors around access to health and social services (Richard et al., 2016), representation in health-related research (Ellard-Gray et al., 2015) and utilization of health technologies (Thurber et al., 2013).

2.3.3. Description of health equity considerations

Integrating, reporting, and identifying equity considerations in health-related research can be challenging because of the various parameters that characterize health equity. Studies suggested strategies for the integration, reporting and identification of equity considerations in health-related research such as clinical trials (Mbuagbaw et al., 2017), clinical guidelines (V. A. Welch et al., 2017b); and HTA(Benkhalti et al., 2021; Culyer & Bombard, 2012).

Research gaps: Researchers also highlighted the importance of assessing the implications of considering health equity in HTA for health systems (Allen, Liberti et al., 2017; Culyer & Bombard, 2012; Hoch et al., 2021). Researchers developed tools to incorporate and report health equity considerations in HTA. For example, (Benkhalti et al., 2021) proposed an equity checklist for health technology assessment (ECHTA) that helped HTA analysts assess potential inequities surrounding a health technology implemented in Quebec, Canada. These authors also advised HTA analysts to explicitly report on health equity analysis conducted as part of HTA. However, studies did not identify criteria for designating HTA recommendations as equity-focused.

Characterizing health equity factors reported in HTA recommendations can help draw attention to the need to systematically conduct health equity analysis in HTA research (Benkhalti et al., 2021; Hosking et al., 2019; Prady et al., 2018). Some researchers have attempted to develop search filters for identifying equity-focused studies (Hosking et al., 2019; Prady et al., 2018). Additional to the parameters described in the PROGRESS-Plus framework for characterizing population groups, researchers advised looking for non-specific terms that suggest group comparison, such as "risk factors for," "were high/low" "at higher risk for" "significant among," and "differed by" (Benkhalti et al., 2021; Hosking et al., 2019; Prady et al., 2018).

For example, the studies identified criteria to characterize equity considerations in HTA. These may include availability or lack of alternative options, applicability according to age and gender, impact on cost and outcomes for individuals not using the technology, variation in effectiveness related to comorbidities and co-medications, literacy level, and differential access due to socioeconomic position (Benkhalti et al., 2021; Culyer & Bombard, 2012; Hoch et al., 2021). Combining specific terms to label social determinants of health and non-specific terms to differentiate the sociodemographic characteristics of research participants can help identify equity-focused studies more accurately.

2.3.4. Patient engagement and equity-focused HTA recommendations

HTA organizations increasingly involve patients in examining health technologies (Facey Helle et al., 2017; Finset, 2017; Moreira, 2015; J. Wale et al., 2017). Patient engagement is increasingly widely accepted as a practical approach for incorporating patient perspectives in developing recommendations to assist decision-makers in their appraisals of health technologies (Bellemare et al., 2018; Facey Helle et al., 2017; O'Flaherty et al., 2021). Sometimes, HTA analysts must engage patients in HTA (Haerry et al., 2018). In HTA, studies reported that patient engagement is essential for capturing patients' perspectives on health technologies (Douglas et al., 2015; Facey Helle et al., 2017; Hicks et al., 2014). The effectiveness of health technologies can be valued differently depending on patients' experiences and preferences (Douglas et al., 2015; Facey, 2019; Hicks et al., 2014). Research also highlighted patient engagement's role in understanding the effectiveness of implemented health technologies (Facey et al., 2018; Press & Richards, 2015; Staley, 2015). Studies reported how differences in perceptions about the meaning of health and experiences with society could shape the framing of health problems among population groups (Brüssow, 2013; Huber et al., 2011).

Health providers and researchers may have limited knowledge of cultural and social differences underpinning perceptions about health problems among those affected (Charlier et al., 2017; Huber et al., 2011; Misselbrook, 2014). For instance, biomedical and public health researchers have extensively used the World Health Organization (WHO) conceptualization of health in their work (Callahan, 1973). WHO defines health as "a state of complete physical, mental and social well-being, and not merely the absence of disease or infirmity" (WHO,1946)(Callahan, 1973). However, several critics have called for a revision of this conceptualization of health because it does not consider the dynamic nature of people's social, technological and environmental challenges (Brüssow, 2013; Charlier et al., 2017; Huber et al., 2011; Misselbrook, 2014). On the other hand, there is no consensus on a new definition to replace the WHO definition of health (Brüssow, 2013; Charlier et al., 2017; Misselbrook, 2014).

Nevertheless, differences between researchers and patients regarding the meaning of health can raise multiple challenges. For example, those differences could negatively impact identifying health problems and access to health technologies (Dipankui et al., 2015; Staley & Doherty, 2016; J. Wale et al., 2017). Studies also described the role of patient engagement in clarifying patients' preferences and values in HTA (Huls et al., 2019; Muhlbacher & Johnson, 2017). Studies identified examples of outcomes of patient engagement in HTA in Australia, Canada, the United Kingdom and the United States of America (Forsythe et al., 2019; Lopes et al., 2016; Manafo et al., 2018; Staley & Doherty, 2016).

Research gaps: More research is, however, needed to elucidate how patient engagement strategies impact the formulation of HTA recommendations (Cavaller-Bellaubi et al., 2021;

Crocker et al., 2017; Facey, 2019; Huls et al., 2019). Recent evidence suggests that patient engagement can help identify relevant health outcomes, address gaps in the evidence on clinical and cost-effectiveness, and assess the local context in HTA recommendations (Cavaller-Bellaubi et al., 2021; Facey Helle et al., 2017; Hashem et al., 2018; Lambert et al., 2018; Rycroft-Malone et al., 2016). It is also important to acknowledge that researchers have used multiple strategies to incorporate equity factors in HTA recommendations (Benkhalti et al., 2021; Busse et al., 2015; Johri & OF, 2012; Marsh & Le, 2015).

For example, the significance and the weight of strategies regarding equity considerations in HTA recommendations will vary depending on the governance system (Espinoza & Cabieses, 2014; Hill & Olson, 2014; Mohara et al., 2012; O'Rourke et al., 2020). Studies identified patient engagement as only one approach to developing equity-focused HTA recommendations (Douglas et al., 2015; Facey Helle et al., 2017; Press & Richards, 2015; Snow et al., 2018). Clarifying the relationships between patient engagement and the development of HTA recommendations can reinforce the validity of recommendations on implementing health technologies and health-related policies (Dipankui et al., 2015; Staley & Doherty, 2016; J. Wale et al., 2017).

Overview of patient engagement approaches in HTA

2.3.5. Definition of patient engagement

In health research, patient engagement involves a partnership between researchers and patients to capture patients' experiences and perspectives to influence health research narratives (Facey et al., 2018; Frank et al., 2020; Higgins et al., 2017). In the literature on patient engagement in HTA, the term patient is used as an umbrella concept to describe the perspectives of any individual living with a health condition, as well as those of caregivers of individuals living with a health condition, patient advocates who support people living with a specific health condition, patient experts who have experience with and technical knowledge about a particular health condition and patient organizations that represent the collective voice of individuals living with a health condition (Higgins et al., 2017; Maguire & Britten, 2017).

2.3.6. Overarching models of patient engagement

Studies reported the importance of active and meaningful engagement, whether patients or the public are involved in HTA processes (Boudes et al., 2018; Cavaller-Bellaubi et al., 2021; de Wit et al., 2020; Facey, 2019; Frank et al., 2020; Snow et al., 2018). Knaapan and Lehoux (2016) have identified three main engagement models in the patient and public involvement literature (Knaapen & Lehoux, 2016). These consist of the *Consumer Choice model,* the *Democratic Voice model,* and the *Lay Expertise model* (Knaapen & Lehoux, 2016). In the Consumer Choice model, patients are consumers of healthcare, with rights of access to information and autonomous use of healthcare. As such, their engagement should enable informed decisions and facilitate adapting health services to their needs and ability to use them (Knaapen & Lehoux, 2016).

The Democratic Voice model is based on the rights of the public to participate in policy-making. It seeks to integrate collective values in policies through the involvement of "ordinary citizens," whose stakes lie in the informed use of healthcare and allocating public resources (Knaapen & Lehoux, 2016). Engagement through that lens seeks to determine the relative value of health technologies across several conditions (Knaapen & Lehoux, 2016; Safaei, 2015a). Lastly, the *Lay Expertise* model describes patients as "experts" in their illness because of the unique knowledge they gain through their experiences with the condition (Facey Helle et al., 2017; Grande et al., 2014; Knaapen & Lehoux, 2016). Engagement based on the Lay Expertise model sought to integrate experiential knowledge and scientific evidence to adopt policies for patients' needs and lived experiences (Facey Helle et al., 2017; Knaapen & Lehoux, 2016).

Overlapping and divergent elements were identified in these models (Knaapen & Lehoux, 2016). For instance, *Consumer Choice*, *Democratic Voice* and *Lay Expertise* all consider aspects of experiences with technologies as lived experiences depend on the ability to use as well as access to and knowledge about the technology (Knaapen & Lehoux, 2016; Maguire & Britten, 2017; Safaei, 2015a). On the other hand, the Democratic Voice Model may lead to discriminatory decisions toward people suffering from rare diseases that require expensive treatment (Knaapen & Lehoux, 2016; Safaei, 2015a). The majority may think it is unfair to them to allocate funds that could treat one hundred citizens to care for only one person, for example (Safaei, 2015a).

While reasons for considering patients' perspectives vary across HTA organizations, advocacy for engaging patients in HTA mainly combines the above models (Chudyk et al., 2022; Facey et al., 2018; Safaei, 2015a; Whitty & Littlejohns, 2015). For example, HTA organizations where people debate democratic rights may focus on implementing patient engagement strategies that consider the diverse representation and meaningful participation of patients to elucidate choices, utilization and fair distribution of health technologies (Bellemare et al., 2018; Cookson & Mirelman, 2017; Lopes et al., 2016; Manafo et al., 2018). In other political systems, HTA organizations may focus their patient engagement strategies on establishing consensus around the use and coverage of health technologies (Coulter, 2012; Espinoza & Cabieses, 2014; Shams Moattar et al., 2016).

2.3.7. Procedural parameters in patient engagement strategies

Patient engagement strategies vary with the context of HTA organizations and the characteristics of the health technology (Cavaller-Bellaubi et al., 2021; Facey Helle et al., 2017; Wahlster et al., 2017a). A uniform taxonomy to describe patient engagement methods has not been reached (Bombard et al., 2018a; Chudyk et al., 2022; Dipankui et al., 2015; Greenhalgh et al., 2019; Manafo et al., 2018). Studies have shown that methods of engaging patients in research vary widely with the stage and context of research (Bombard et al., 2018a; Concannon et al., 2012; Facey Helle et al., 2017; Frank et al., 2020). Processes of patient engagement in health-related research and HTA are increasingly gaining researchers' attention (Biddle et al., 2021; Chudyk et al., 2022; Davis et al., 2019; de Wit et al., 2020; Facey, 2019; Frank et al., 2020; A. Sharma et al., 2018).

Research gaps: A recent scoping review identified 14 models and frameworks of patient engagement in health-related research with 18 overlapping and 57 divergent elements across six conceptual areas (Chudyk et al., 2022). Considering this conceptual landscape, it becomes crucial to identify which procedural elements can support the purpose of patient engagement (Chudyk et al., 2022; de Wit et al., 2020; Frank et al., 2020; Greenhalgh et al., 2019).

Table *2* illustrates the procedural parameters I considered in the review of patient engagement strategies.

1. Types of engagement: Patient engagement in HTA involves partnerships between HTA institutions and individual patients or patient groups (Facey Helle et al., 2017; Manafo et al., 2018; Wahlster et al., 2017a). In a direct engagement, researchers talk directly to patients, whereas a patient representative works with researchers on behalf of members of patient groups (Facey Helle et al., 2017; Manafo et al., 2018; Wahlster et al., 2017). The primary purpose of

patient groups is to ensure that patient perspectives are reflected in decisions regarding their health (Hicks et al., 2014; Maguire & Britten, 2017; Mercer et al., 2020). In the context of HTA, their work covers a wide range of interests, including concerns related to effectiveness, legal and ethical issues, accessibility, and pricing of health technologies (Cavaller-Bellaubi et al., 2021; Facey, 2019; Manafo et al., 2018; Mercer et al., 2020).

Table 2: Procedural parameters in patient engagement.

Parameters of patient engagement processes		Some relevant sources
1. Types of engagement	Direct; indirect	Facey Helle et al., 2017; Manafo et al., 2018;
2. Levels of engagement	Information; consultation; Co-production	Frank et al., 2020; Wortley et al., 2017; Dipankui et al., 2015; Domecq et al., 2014;
3. Patients' roles	Co-investigators; members of an advisory committee, staff, key informants, expert panels	Biddle et al., 2021; Concannon et al., 2012; Concannon et al., 2019; Wortley et al., 2017;
4. Models of decision-making	Vote; consensus	Manafo et al., 2018; Wahlster et al., 2017; Concannon et al., 2019; Wortley et al., 2017
5. Modes of engagement	Interview, focus groups, survey, written submission	Biddle et al., 2021; Manafo et al., 2018; Concannon et al., 2019; Wortley et al., 2017
6. Modalities of engagement	In-person; digital technologies (e.g., phones, web portal, survey platforms, mail meeting apps)	Cordoş et al., 2017; Jenssen et al., 2016; Liddy et al., 2017; Concannon et al., 2019;

Patient groups are generally member-based organizations composed of individuals who share everyday experiences with a health condition or a disease (Facey Helle et al., 2017; Maguire & Britten, 2017; Manafo et al., 2018). Patient groups can be locally grown or branches of bigger international and national patient advocacy groups(Facey Helle et al., 2017; Maguire & Britten, 2017). It is important to note that advocacy approaches between patients and public interest groups may differ in HTA. Public interest groups usually have a similar purpose as patient groups, yet they tend to focus more on finding balance in using public money to fund health technologies (Facey Helle et al., 2017; McCoy et al., 2019; Moreira, 2015; Warsh, 2014). Other public interest groups may be more preoccupied with the representation of the societal values of the public in HTA recommendations(Facey Helle et al., 2017; McCoy et al., 2019; Moreira, 2015; Warsh, 2014).

2. Levels of engagement: When considering patients' influence on research activities, patient engagement may consist of informing patients of the research, consulting patients for input on research activities and co-producing research with patients (Batalden et al., 2016; Facey Helle et al., 2017; Frank et al., 2020; Higgins et al., 2017). Although described in several models as the lowest level of patient engagement (Dipankui et al., 2015; Domecq et al., 2014), some researchers argued that informing patients of the research should not be considered as "engagement" (Frank et al., 2020; Hahn et al., 2017; Jacklin & Kinoshameg, 2008; Snow et al., 2018). At this level, communication is unidirectional -from researcher to patient- and does not imply any expectation of patient feedback (Hahn et al., 2017; Hicks et al., 2014; Jacklin & Kinoshameg, 2008). Also, researchers are not obligated to respond to patient comments (Hahn et al., 2017; Hicks et al., 2014; Jacklin & Kinoshameg, 2008).

However, consultations and co-production provide more significant opportunities for capturing patients' experiences and expertise to leverage the impact of research (Batalden et al., 2016; Facey Helle et al., 2017; Finset, 2017; Frank et al., 2020; Wortley et al., 2017). At those levels, patients must receive sufficient information and support to actively contribute to the research activity (Frank et al., 2020; Goodman & Sanders Thompson, 2017; Wortley et al., 2017). In turn, researchers must acknowledge and demonstrate how researchers value patients' input in decisions (Batalden et al., 2016; Finset, 2017; Frank et al., 2020; Wortley et al., 2017).

3. Patient Roles and Models of Decision-Making: The roles of patients vary with the context and phases of research(Concannon et al., 2012; Domecq et al., 2014; Frank et al., 2020). Patients' roles may consist of co-investigators, members of advisory committees, expert panels, and research participants (Biddle et al., 2021; Concannon et al., 2019; Facey Helle et al., 2017; Frank et al., 2020; Manafo et al., 2018). These roles add value to the research process and benefit patients (Press & Richards, 2015; Rycroft-Malone et al., 2016; Snow et al., 2018; J. L. Wale et al., 2017; Wortley et al., 2017). Studies reported voting and consensus-building as the principal ways for patients to contribute to decisions in research activities (Concannon et al., 2019; Facey Helle et al., 2017; Manafo et al., 2018).

4. Modes and modalities of engagement: Modes and modalities of engagement may change at different phases of the research process. Typically, patients provide input through an interview, survey, focus group or written submission (Concannon et al., 2019; Facey Helle et al., 2017; Manafo et al., 2018). Researchers often collect patient input either in-person or through digital technologies such as phones, web portals, survey platforms, social media and virtual meeting applications (Catarinella & Bos, 2016; Cordoş et al., 2017; Jenssen et al., 2016; Liddy et al., 2017).

2.4. Characteristics of diverse and inclusive patient engagement

2.4.1. Definition of inclusive patient engagement

Studies described inclusive patient engagement as creating a research environment where research teams manage power dynamics and research processes in ways that enable patients to participate positively and productively (Black et al., 2018; Cavaller-Bellaubi et al., 2021; Khuntia et al., 2022; Reynolds et al., 2021; Shimmin et al., 2017). Some authors posit that inclusive patient engagement can only happen over time, as it should embed enough flexibility to allow patients to participate at their own pace (Cavaller-Bellaubi et al., 2021; Gill et al., 2018; Khuntia et al., 2022). This relationship-building aspect of inclusive patient engagement also acknowledges the importance of patients' lived experiences and all forms of knowledge (Cavaller-Bellaubi et al., 2021; Khuntia et al., 2022; Roche et al., 2020; Shimmin et al., 2017)

2.4.2. Inclusive patient engagement

Another aspect of inclusion in research is demonstrating to patients that their input is valued (Biddle et al., 2021; Cavaller-Bellaubi et al., 2021; de Wit et al., 2020; Manafo et al., 2018; Snow et al., 2018). For example, two recent studies found that allocating a budget to patient engagement activities and providing practical support could help demonstrate patient engagement's value in research (Biddle et al., 2021; Cavaller-Bellaubi et al., 2021).

Practical support for promoting inclusive patient engagement may consist of training patients on research processes, training researchers on the use of lay language, adapting communication tools, and providing travel accommodations (Black et al., 2018; Davis et al., 2019; Dawson et al., 2018; Gunn et al., 2021; Roche et al., 2020). In addition to those enabling factors, evidence shows that patients feel their contribution is valued when researchers consider their input while maintaining rigour in the research process (Black et al., 2018; Chudyk et al., 2022; Gill et al., 2018; Gunn et al., 2021). Inclusive patient engagement also helps patient input in knowledge products such as HTA recommendations (O'Flaherty et al., 2021).

2.4.3. Marginalizing engagement practices

Other studies have, however, suggested that some methods of engaging patients might contribute to further excluding marginalized and underrepresented populations from taking part in research (Bombard et al., 2018a; Crockett et al., 2019; Dawson et al., 2018; Ellard-Gray et al., 2015; Hahn et al., 2017; Shimmin et al., 2017). For example, some marketing-based patient engagement models that use tokenism can discourage some individuals from signing up for research engagement (Bombard et al., 2018a; Hahn et al., 2017; Ocloo & Matthews, 2016). Because these individuals realize that researchers contacted them just for the appearance of engagement, there is no expectation of actual participation or feedback (Bombard et al., 2018a; Hahn et al., 2017; Ocloo & Matthews, 2016).

In other cases, researchers have genuine intent to engage patients from marginalized groups in research, but researchers may structure the engagement in ways that leave patients feeling stigmatized and unwelcomed (Crockett et al., 2019; Dawson et al., 2018; Roche et al., 2020; Shimmin et al., 2017). In those instances, the researchers may not acknowledge certain patients' knowledge and experiences(Dawson et al., 2018; Shimmin et al., 2017); other patients may present themselves as gatekeepers. They control access to patient engagement for others in their communities (Ellard-Gray et al., 2015; Hahn et al., 2017). For those reasons, some researchers have called for applying the health equity and social justice lens to existing patient engagement to help increase diversity and inclusion in patient engagement processes (Dawson et al., 2018; Khuntia et al., 2022; Roche et al., 2020).

2.4.4. Diverse representation in patient engagement

Studies described diversity in patient engagement as "bringing less traditional patients' voices" in health research (Dawson et al., 2018; Khuntia et al., 2022; Roche et al., 2020). These less traditional patients' voices may consist of people who are facing barriers to participating in research because of their living conditions, geographical locations, physical and mental health conditions, socioeconomic status and lower literacy levels (Gill et al., 2018; Reynolds et al., 2021; Roche et al., 2020). Underrepresented population groups in health research may also include people who are avoiding the health systems because of traumatic experiences due to stigma and systemic barriers to accessing healthcare (Davis et al., 2019; Dawson et al., 2018; Krieger, 2014; Nyblade et al., 2019; Roche et al., 2020). Achieving diversity in the representation of population groups comes with challenges related to representativeness (Domecq et al., 2014; Dukhanin et al., 2020; Gill et al., 2018; Rowland & Kumagai, 2018). Other patients may challenge the credibility of representatives that researchers recruit to participate in studies, even when those representatives have profiles that match the research purpose (Eccles et al., 2018; Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018).

Researchers have resorted to several strategies to address representativeness in patient engagement. Some researchers have used democratic rights as the guiding principle for engaging patients in studies (Fischer & Van de Bovenkamp, 2019). A patient representative is, therefore, selected through either nomination or election to contribute to patient engagement on behalf of other patients (Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018). Some authors argue that this type of representation often poses issues surrounding accountability, authorization and political representativeness (Dukhanin et al., 2020; Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018; Safaei, 2015a).

Another approach is to consider statistical representation, which implies that a patient can relay the average experience of all patients with the same condition (Rowland & Kumagai, 2018). An obvious issue with this approach is the assumption that the selected patient representative has the skills to convey the experiences of the whole patient group (Crockett et al., 2019; Mazor et al., 2016; Rowland & Kumagai, 2018). Other researchers have used symbolic representation, which focuses on patient's unique experiences to bring individual perspectives into constructing research narratives (Dukhanin et al., 2020; Rowland & Kumagai, 2018). From this viewpoint, a patient representative shares their story as their own authentic and unique experience, which may not necessarily reflect the lived experiences of other people who may have experienced the same condition (De Santis et al., 2019; Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018).

Research gaps: Based on the challenges discussed above, it is equally important to promote diversity of perspectives regardless of the rationale used to recruit patients (De Santis et al., 2019; Reynolds et al., 2021; Snow et al., 2018). Research organizations must equip researchers and patients to ensure patient engagement activities acknowledge differences in knowledge, skills and interests (Black et al., 2018; Crockett et al., 2019; Hahn et al., 2017). Acknowledging differences among patient participants and researchers can lead to diverse perspectives and a new understanding of health outcomes (Charlier et al., 2017; Reynolds et al., 2021; Roche et al., 2020). For example, the level of skills and comfort to participate in engagement activities from both patients and researchers can influence the types of perspectives generated through differences that emerge in patient engagement (Crockett et al., 2019; Dawson et al., 2018; Reynolds et al., 2021; Snow et al., 2018).

2.5. Digital technologies for diverse and inclusive patient engagement

2.5.1. Definition of digital technologies

Digital technologies consist of all electronic technologies (e-technologies) and applications that use information in numeric code (Aziz & Madani, 2015). Digital technologies can support interactive and non-interactive functions through the internet and the web, which sustain the networking of computers and individuals (Aziz & Madani, 2015; P. Sharma & Kaur, 2017). Digital technologies cover various information and communication technologies to support data production, management, and processing (Aziz & Madani, 2015; Mitchell & Kan, 2019).

2.5.2. Use of digital technologies in population health

Digital technologies can enable social interactions, facilitate access to resources and asynchronously enhance the sharing of information using various levels of interactions (Aziz & Madani, 2015; Mitchell & Kan, 2019). Digital technologies also provide opportunities to collect, manage and analyze large amounts of data, allowing building patterns characterizing users (Aziz & Madani, 2015). Examples of applications of digital technologies comprise a telephone, intelligent devices such as smartphones, iPads, and digital watches; smart devices for persons with disabilities such as assisted devices for impaired vision, impaired speech and reduced mobility; social media outlets such as Facebook and Twitter, blogs, websites, data management and webinar software (Aziz & Madani, 2015).

In addition, digital technologies improve the capacity for conducting and implementing research to inform the population and public health practice (Brewer et al., 2020; A. Sharma et al., 2018). In population and public health, digital technologies help deliver services to the public (Aziz & Madani, 2015; Mitchell & Kan, 2019; Ting et al., 2020); connect community members (Kent & Yellowlees, 2015); support professional training of healthcare providers (Jackson et al., 2018; Lawn et al., 2017), and promote public health (Petkovic et al., 2021b).

Studies have also described digital technology functions for reducing constraints related to locations, disabilities and communication needs (Brewer et al., 2020; Mitchell & Kan, 2019; Ting et al., 2020). Some digital technologies, like digital vaccine passports, can enable patients to use health technologies by helping users access information, manage immunization records, and share lived experiences while collecting data on vaccination uptake (Wilson & Flood, 2021).

However, research on assessing digital technologies to support diverse and inclusive patient engagement in health-related research remains limited.

Another example is using various digital technologies to mitigate the impact of public health threats like the novel coronavirus disease 2019 (COVID-19)[1] through facilitating monitoring, surveillance, detection, and prevention (Krausz et al., 2020; Ting et al., 2020). COVID-19 is a highly transmissible disease with moderate to severe clinical outcomes, a disproportionate burden, and those living in vulnerable social conditions (Khalili et al., 2020). The COVID-19 pandemic has also revealed the fragility of health systems worldwide, thereby creating an urgency to adapt existing processes of organization and provision of healthcare, including harnessing the potential of digital technologies for such purposes (Krausz et al., 2020; Ting et al., 2020).

The COVID-19 pandemic has also caused impactful societal and economic disruption (Giuntella et al., 2021), reshaping social interactions and access to services (Ayouni et al., 2021; Giuntella et al., 2021; Krausz et al., 2020; Ndaïrou et al., 2020). In adapting to this new reality, online social interactions and virtual service delivery helped reduce social isolation while limiting physical contact (Giuntella et al., 2021; Ohannessian et al., 2020; Ting et al., 2020). In addition to the burden of the disease on specific populations (Khalili et al., 2020; Wang & Tang, 2020), the health equity impact of the response to COVID-19 is widely discussed. For instance, some authors are positioning digital inequalities as a social determinant of health because of the central role digital technologies play in implementing surveillance strategies (e.g. contact

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[1] WHO declared a public health emergency at the end of January 2020 after reports of an outbreak of a pneumonia of unknown origin was reported in China on December 2019(Khalili et al., 2020, p. 19). The name coronavirus disease 2019 (COVID-19) was announced by WHO in February 2020.

tracing), accessing services, and organizing care during the current COVID-19 pandemic (Beaunoyer et al., 2020; Yao et al., 2022).

Institutions envisioning health technology assessment (HTA) as a process leading to health equity must consistently adapt their patient engagement strategies. Equitable patient engagement processes will allow HTA organizations to face challenges that arise from growing recognition of democratic rights (Safaei, 2015a), easy access to information (King et al., 2016; Powell et al., 2003), population mobility (van der Waal et al., 2017), and the sharp increase to online service delivery after the COVID-19 pandemic (Khalili et al., 2020). Improving patient involvement in the HTA process can help better balance experiential and scientific knowledge about the effectiveness of health technologies and contribute to developing more equity-focused HTA recommendations.

Research gaps: As digital technologies continue to transform how patients interact with their environment (Doupi, 2016; Kent & Yellowlees, 2015; Mitchell & Kan, 2019), it becomes essential to explore how its potential can leverage the inclusion and participation of patients from diverse backgrounds in the HTA process. Furthermore, patients are demanding more accountability from health authorities and calling for considering their voices in decisions regarding their health (Biddle et al., 2021; Crockett et al., 2019; de Wit et al., 2020; Snow et al., 2018; Staley, 2015). Digital technologies offer opportunities for implementing diverse and inclusive patient engagement strategies (Cordoş et al., 2017; Jenssen et al., 2016; Mitchell & Kan, 2019), which can enhance the development of equity-focused HTA recommendations. HTA recommendations inform policies such as drug coverage, healthcare services, preventive interventions, and public health workforce training, all of which have health equity implications when rolled out to the public.

2.6. Discussion

The literature review helped identify critical strategies for incorporating health equity considerations in developing HTA recommendations. HTA was discussed as a policy analysis framework to help develop recommendations for informing decision-makers on the justification for funding or not a health technology. Studies reported that patient engagement is critical in identifying health equity factors for HTA recommendations. Health technologies could be valued differently based on socially constructed characteristics of population groups, types of interventions and features of the disease. Framing patient engagement as an intervention with specific input, output, and effect on developing HTA recommendations can help better evaluate patient engagement strategies.

The studies showed that engagement models for developing HTA recommendations that patient engagement is essential in the health equity analysis for HTA. The main output of patient engagement activities consists of a report on patient preferences and values. The review also identified conceptual frameworks for incorporating health equity considerations in HTA recommendations. The review did not identify a uniform taxonomy for characterizing patient engagement strategies. I did not find studies that quantitatively described the influence of patient engagement on integrating health equity factors in HTA recommendations.

Digital technologies can help with identifying, recruiting, and engaging diverse patients. However, none of the included studies assessed the impact of patient engagement on health equity analysis in HTA. Some authors have highlighted the importance of further investigating these topics to help enhance the impact of health research on improving health outcomes. Future research could aim to determine the characteristics of patient engagement strategies in health-related research, including HTA, identify resources to implement patient strategies, describe the characteristics of digital technologies to support diverse and inclusive patient engagement and assess the impact of patient engagement on the development of equity-focused HTA recommendations.

2.8.1. Conclusion

The ultimate purpose of HTA recommendations is to provide a scientific base to decision-makers to inform their decision on funding health technologies while considering the budget impact and the complexity of maintaining the availability and use of the health technology under review. Understandably, it might be challenging to isolate the effect of patient engagement as one pillar of the evidence used to inform such a complex decision. Therefore, it is essential to explore how current patient engagement strategies could be improved to increase their weight in formulating equity-focused HTA recommendations.

Chapter 3: Methodology

3.1. Overview of Chapter 3

In Chapter 3, I present a brief overview of the theoretical approach and study design of the dissertation. I have already explained my philosophical paradigm in Chapter 1. Also note that each manuscript has a detailed methods section.

3.2. Theoretical approach: Knowledge-to-action (KTA) cycle

I used the knowledge-to-action (KTA) cycle as an overarching theoretical approach to guide the implementation of the research project. I selected the KTA cycle because HTA is a knowledge creation process where the knowledge product is HTA recommendations. HTA recommendations must reflect the needs and interests of multiple stakeholders, including patients and decision-makers (Stevens et al., 2003; Straus et al., 2013). The KTA cycle provides a systematic approach to developing knowledge that can affect organizational changes and health outcomes (Straus et al., 2013).

The KTA comprises two distinct yet complementary phases: the knowledge creation and the action cycle phase, realized in three and seven steps, respectively (Graham et al., 2006). Researchers can use parts of the KTA cycle to support their work in any order (Graham et al., 2006; Straus et al., 2009). **Figure 4** demonstrates the application of the KTA cycle to the thesis, which includes the following steps: *evaluate outcomes, determine the know/do gap, and assess barriers and facilitators to knowledge use.*

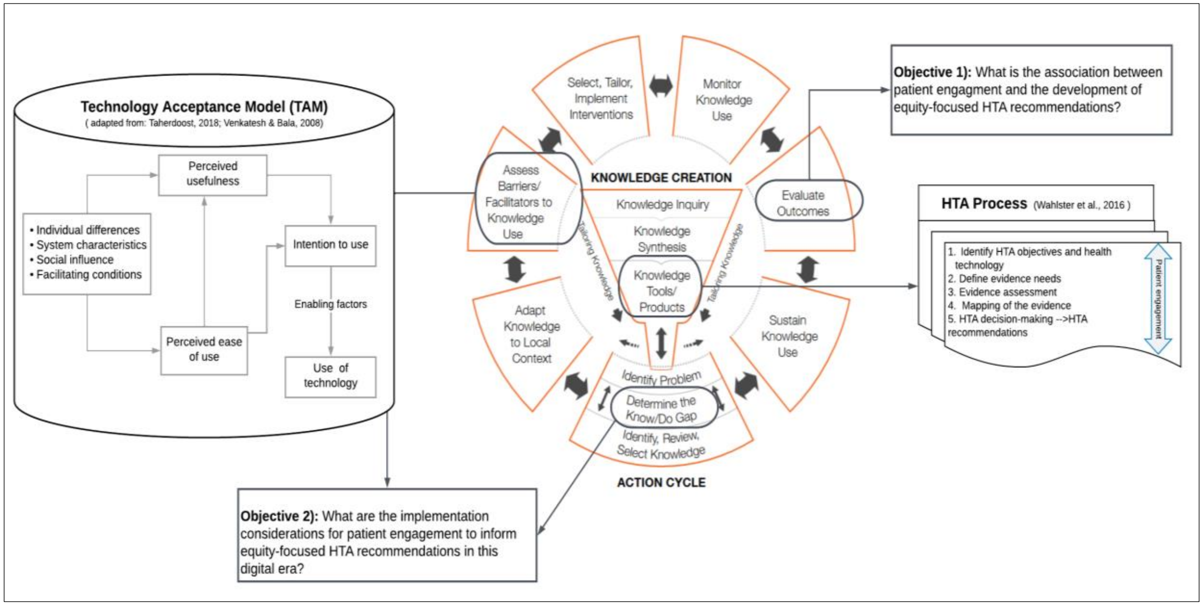

Figure 4: Application of KTA cycle to the thesis (Adapted from Graham et al. 2006).

3.2.1. Evaluate outcomes

During this step, researchers seek to understand the relationships between the intervention -*knowledge*- and changes in the outcome (Graham et al., 2006). In the context of this project, the outcome of interest is equity-focused HTA recommendations and the intervention of patient engagement. I used 60 HTA reports from CADTH and HQO to evaluate the relationships between patient engagement processes and the development of equity-focused HTA recommendations.

3.2.2. Determine the Know/Do Gap

In this step, researchers and stakeholders work together to identify a practice problem, identify evidence-based solutions, and formulate recommendations to address the previously identified practice problem (Graham et al., 2006). As shown in **Appendix A**, I consulted three patients and four HTA analysts who had experience with HTA processes in Canada to determine the scope of this dissertation. I also conducted a narrative review to assess the state of evidence on patient engagement processes to inform the development of equity-focused HTA recommendations. The stakeholder consultations and the narrative study revealed a need for more evidence on relationships between patient engagement processes and health equity-focused HTA recommendations. There is also a need to develop more inclusive patient engagement. I discussed the research gaps in Chapters 1 and 2. For this dissertation, the Know/Do gap encompasses implementation considerations for patient engagement that can support the integration of health equity factors in HTA recommendations.

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3.2.3. Assess barriers and facilitators to the use of knowledge

This phase helps identify the factors that can impact the effective use of knowledge to achieve the intended changes (Graham et al., 2006). In the context of this dissertation, it is crucial to describe the drivers that may influence the implementation of patient engagement to inform the development of equity-focused HTA recommendations. These drivers may be inherent in characteristics of HTA, patient engagement strategies and digital technologies. Other planned-action theories can help elucidate those drivers (Graham et al., 2006). For this dissertation, I used a behavioural model, the Technology Acceptance Model (TAM), to help action the knowledge for organizational change (Graham et al., 2006; Taherdoost, 2018).

As illustrated in **Figure 4**, I used the TAM to supplement the KTA for describing barriers and facilitators to using digital technologies in implementing inclusive patient engagement. The TAM provides a theoretical framework to explain the adoption of new technologies. It suggests that using a specific technology depends on multiple parameters, including the features of the technology, social influence, facilitating conditions and the characteristics of the intended users (Taherdoost, 2018; Venkatesh & Bala, 2008). These parameters affect users' perceptions of the technology's usefulness and ease (Taherdoost, 2018). Perceived usefulness is related to the functionality of the technology in helping to solve the identified problem, whereas perceived ease of use concerns efforts required to effectively address the issue at hand (Taherdoost, 2018). Based on the TAM, perceived usefulness and ease of use determine the intention to use a specific technology. Intention to use coupled with enabling conditions such as organizational support will increase the chance of people using the technology (Taherdoost, 2018).

3.3. Research design: explanatory sequential mixed methods

I used an explanatory sequential mixed methods approach to answer the above research questions. This approach uses findings from quantitative data analysis to inform data collection for subsequent qualitative analysis (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017). I selected a mixed methods approach because it can reveal relationships between variables and elucidate the context and meanings of the findings with research participants (Regnault et al., 2018; Shorten & Smith, 2017; Vedel et al., 2019). The qualitative and quantitative results have equal value in helping expand current evidence on the topic covered (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017).

Another reason for selecting the above research design is the lack of evidence on relationships between patient engagement and equity in HTA (Facey et al., 2018; Huls et al., 2019; Mitchell & Kan, 2019). Using multiple data sources such as literature, consultations with stakeholders, and review of institutional reports can help elucidate the context (Abelson et al., 2016; Biddle et al., 2021; Facey et al., 2018). As illustrated in Figure 2, I analyzed each study separately and used the results in an integrated discussion to answer research questions (Creswell & Clark, 2017; Schoonenboom & Johnson, 2017).

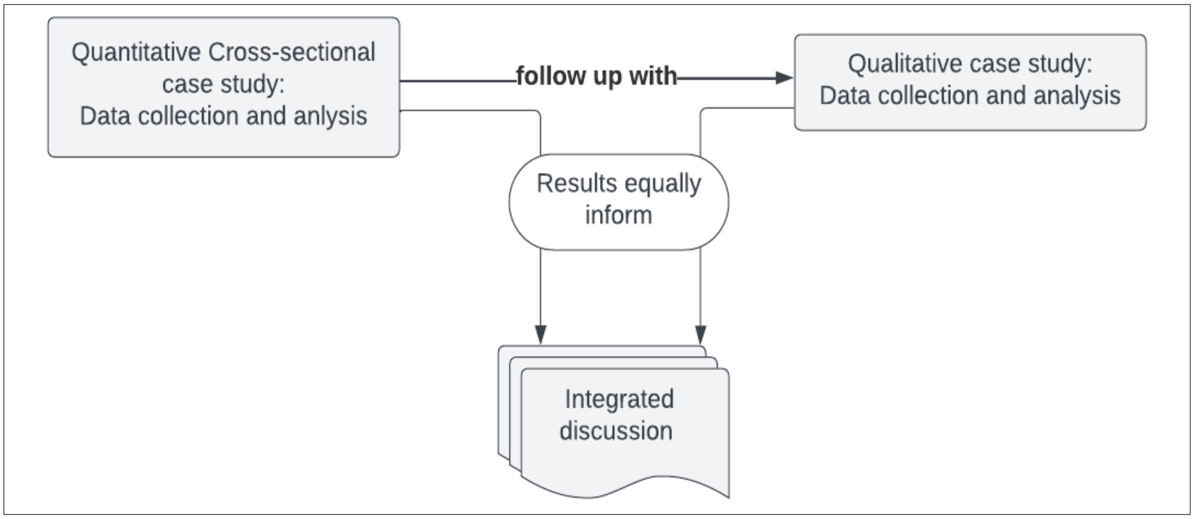

Figure 2: Explanatory sequential mixed methods design to the dissertation.

Chapter 4: Results

4.1. Overview of Chapter 4

The results of the dissertations are discussed in Chapters 5,6 and 7. The first manuscript, Chapter 5, covers the first objective of the dissertation. It is a cross-sectional explanatory case study that examines the relationships between patient engagement and equity-focused HTA recommendations. The second and third manuscripts are qualitative explanatory case studies. They address the second objective of the dissertation, which was to identify implementation considerations for patient engagement in HTA. Chapter 6 describes findings from patients on the second research objective. Chapter 7 describes the additional results of the qualitative case study from HTA analysts' perspectives on the second research objective.

Chapter 5: Manuscript 1

Title: Patient engagement and equity-focused health technology assessment (HTA) recommendations: a cross-sectional case study

5.1. Overview of Chapter 5

Chapter 5 is a complete manuscript describing the dissertation's quantitative portion. The manuscript covers the first objective of the dissertation about the examination of the relationships between patient engagement processes and the development of equity-focused HTA recommendations. Chapter 5 contains the following sections: abstract, background, methods, results, and discussion.

Targeted journal: International Journal of Technology Assessment in Health Care (IJTAHC)

Authors’ contribution:

- Rosiane Simeon (RS) is the Ph.D. candidate and lead author of the manuscript. Rosiane led the planning, research design, data collection, analysis, and manuscript writing.
- Peter Tugwell and Vivian A. Welch co-supervised the Ph.D. candidate and contributed to the planning, research design and review of the manuscript.
- Ian D. Graham, Kumanan Wilson, Reiner Banken, Shehzad Ali, and Janet H. Roberts are the Thesis Advisory Committee (TAC) members and contributed to the research design and review of the manuscript.
- RS will confirm all co-authors before publication.

5.2. Abstract

Background: Health technology assessment (HTA) is a form of policy analysis that informs decisions about funding and scaling up health technologies to improve health outcomes. An equity-focused HTA recommendation explicitly addresses the impact of health technologies on individuals disadvantaged in society because of specific health needs or social conditions. However, there is a lack of evidence on the relationships between patient engagement processes and the development of equity-focused HTA recommendations.

Objectives: To assess relationships between patient engagement processes and the development of equity-focused HTA recommendations.

Methods: We analyzed 60 HTA reports published between 2013 and 2021 in two Canadian organizations: the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO).

Results: Quantitative analysis of the HTA reports showed that direct patient engagement (OR: 3.85; 95% CI: [2.40 – 6.20]) and consensus in decision-making (OR: 2.27; 95% CI: [1.35 – 3.84]) were more likely to be associated with the development of equity-focused HTA recommendations in comparison to indirect patient engagement (OR: 0.26; 95% CI: [0.16 – 0.41]) and voting (OR: 0.44; 95% CI: [0.26 – 0.73]).

Conclusion: The results can inform the designing of patient engagement strategies in HTA. The findings have implications for practice, research, and policy.

5.3. Background

Decision-makers can achieve health equity by acting on modifiable factors that cause unfair distribution of health outcomes across populations (Braveman & Gruskin, 2003; Marmot, 2017). Researchers proposed various tools to facilitate such modifications to pursue health equity, including knowledge production (Tugwell et al., 2006), practice guidelines (Welch et al., 2017), and policy analysis (Benkhalti et al., 2021). Health technology assessment (HTA) is a form of policy analysis that informs decisions about funding and scaling up health technologies (O’Rourke et al., 2020; Stevens et al., 2003). Health technologies are inherent in health service infrastructure and include pharmaceuticals and non-pharmaceutical products such as diagnostic, preventive, treatment and rehabilitation procedures used to support health and well-being (O’Rourke et al., 2020; Stevens et al., 2003). Organizations, such as the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO), develop HTA recommendations by examining available evidence on health technologies' cost-effectiveness and safety (O'Rourke al., 2020; Stevens et al., 2003).

An equity-focused HTA recommendation explicitly addresses the impact of health technologies on individuals disadvantaged in society because of specific health needs and social characteristics (Benkhalti et al., 2021; Braveman & Gruskin, 2003; Tugwell et al., 2006b). As such, a health equity-focused recommendation puts those with identified higher vulnerability risks at the center of the HTA issue (V. Welch et al., 2008). Studies identified the disparate and inequitable distribution of health outcomes based on socially constructed characteristics such as those described in the PROGRESS-Plus framework (O’Neill et al., 2014). PROGRESS stands for Place of residence, Race/ ethnicity, Occupation, Gender, Religion, Education, Socioeconomic status, and Social capital. The Plus designates other reported strata, such as sexual orientation and individuals with disabilities, used to describe participants in health-related research and interventions (O'Neill et al., 2014).

Patient engagement involves a partnership between researchers and patients to capture patient perspectives to influence knowledge creation, like HTA recommendations (Facey Helle et al., 2017; Frank et al., 2020; Higgins et al., 2017). HTA organizations increasingly adopt patient engagement as a practical approach for incorporating patient perspectives in developing recommendations (Bellemare et al., 2018; O'Flaherty et al., 2021). Some HTA organizations have made patient engagement mandatory in their HTA process (Haerry et al., 2018). In practice, HTA analysts engage individual patients or patient groups to inform their work (Facey Helle et al., 2017; Manafo et al., 2018). Evidence is growing on the importance of patient engagement in HTA (Bidonde et al., 2021; Facey et al., 2018; Scott et al., 2017). However, there is a need for more research that examines the relationships between patient engagement processes and the incorporation of equity considerations in HTA recommendations (de Wit et al., 2020; Facey, 2019; Mercer et al., 2020).

It is essential to acknowledge that multiple strategies can help incorporate equity factors in HTA recommendations (Benkhalti et al., 2021; Busse et al., 2015). For example, the significance and the weight of strategies regarding equity considerations in HTA recommendations will vary depending on governance systems (Espinoza & Cabieses, 2014; Hill & Olson, 2014; Mohara et al., 2012). Patient engagement is only one approach to developing equity-focused HTA recommendations (Cookson et al., 2017; Snow et al., 2018). Clarifying the relationships between patient engagement and the integration of health equity factors in HTA recommendations can reinforce the validity of those recommendations. Decision-makers use HTA recommendations to inform policies such as drug coverage, healthcare services, preventive interventions, and public health workforce training, all of which have equity implications when rolled out to the public.

5.4. Objectives

The study examines the association between patient engagement processes and the development of equity-focused HTA recommendations. We addressed the following research questions to achieve the objective:

1. What are the characteristics of patient engagement processes in HTA?
2. What are the characteristics of equity-focused HTA recommendations?
3. What patient engagement processes are associated with equity-focused HTA recommendations?

5.5. Methods

5.5.1. Study design

We used a cross-sectional case study design to assess the prevalence of equity-focused HTA recommendations and to determine the relationships between patient engagement processes and equity-focused HTA recommendations using a sample of 60 reports from two Canadian HTA organizations. Case studies help generate an in-depth understanding of a complex issue in its natural setting (Carey et al., 2012; Crowe et al., 2011a). The case here consists of patient engagement processes for HTA within the Canadian context. We decided to use an explanatory case study approach because it can help generate theories about the influence of patient engagement processes on incorporating equity factors in recommendations based on the context of HTA in Canada

We calculated the sample size based on adequacy for logistic regression(Jenkins & Quintana-Ascencio, 2020; Melsen et al., 2014). We used an earlier study that analyzed equity factors in 19 HTA agencies(Busse et al., 2015) The study found that around 50% of the HTA agencies considered equity factors through their methods or analysis of legal and ethical issues. Also, another study that examined equity considerations in the World Health Organization (WHO) guidelines showed that only 25% of the guidelines contained PROGRESS-Plus items (Dewidar et al., 2020).

We expected more equity-focused recommendations in the HTA reports than in WHO guidelines. So, we used a ratio of 40%, which gave a sample size of 50. We used 60 reports to be more cautious. We expected HTA to contain more equity factors than WHO guidelines because HTA must consider the local context in their analysis of health technologies in comparison to WHO guidelines that require further adjustment before their implementation in a country. In this study, we also described the processes of patient engagement and the characteristics of equity-focused HTA recommendations.

5.5.2. *Identification of reports: inclusion and exclusion criteria*

We used a purposeful sample of 60 HTA reports that contained patient input published between 2013 and 2021 from CADTH and HQO. We only considered HTA organizations that used patient engagement in their HTA methodologies. We decided on Canadian reports because research showed that HTA reviews are context-bound, and patient engagement varied greatly across organizations and within countries (Allen, Liberti, et al., 2017; Manafo et al., 2018; Vreman et al., 2020). Selecting HTA reports with patient engagement strategies in contexts that are not comparable could affect the accuracy of the study findings and their generalizability(Jenkins & Quintana-Ascencio, 2020; Melsen et al., 2014).

While Canada has universal coverage, healthcare delivery remains a provincial and territorial responsibility (Martin et al., 2018). Provinces and territories are responsible for implementing HTA recommendations in Canada (Martin et al., 2018). So, we decided to select a purposive sample of HTA reports from organizations operating at the provincial and federal levels to help understand how their respective patient engagement strategies may have impacted the development of equity-focused HTA recommendations authorities.

For this explanatory case study, we focused on HTA reports from CADTH and HQO after searching the International Network of Agencies for Health Technology Assessment (INHATA) database (INAHTA) [Internet], 2021). We also scanned the websites of five health ministries in Canada and looked for publicly available HTA reports that used patient engagement published in English. CADTH and HQO’s HTA reports are publicly available and have all the information needed to complete the study. In addition, we selected CADTH for applying its HTA products to the federal jurisdiction and HQO for applying their HTA recommendations to the provincial jurisdiction only.

We defined patient engagement as a series of activities allowing two-way communication between patients and HTA organizations that enable patients to share feedback on health technologies and to participate in decision-making on informing HTA recommendations (Bombard et al., 2018b; Facey Helle et al., 2017; Frank et al., 2020). Patient engagement may be direct when HTA analysts consult patients to collect their input (Patient Engagement Action Team, 2017). Indirect patient engagement involves HTA organizations working with patient groups to gather feedback on health technologies (MacPhail & Shea, 2017).

The outcome of interest for this study was an equity-focused HTA recommendation. We defined equity-focused HTA recommendation as a recommendation containing at least one PROGRESS-Plus item in the rationale and scientific evidence to support HTA funding decisions (Benkhalti et al., 2021; Busse et al., 2015).

We selected reports based on types of HTA review, years of publications as well as methods of patient engagement. Contrary to HQO, which did not categorize HTA products on its website, CADTH had several HTA products with specific patient engagement methodologies. As shown in **Table 3**, we selected reports from the following HTA products from CADTH: Common Drug Review (CDR) and pan Canadian Oncology Drug Review (pCODR).

Table 3: Characteristics of included reports.

Characteristics	Description	n (%)
Year of publications		
2013-2015	Earlier implementation period	9 (15%)
2016-2021	Recent implementation period	51(85%)
Types of HTA review		
pan-Canadian Oncology Drug Review (pCODR)	CADTH reports focused on cancer drugs	15 (25%)
Common Drug Review (CDR)	CADTH reports focused on non-cancer drugs	25 (42%)
Health Quality Ontario (HQO)	HQO reports	20 (33%)

We considered HTA reports with at least one of the following factors: common drugs, oncology drugs, medical devices, and virtually delivered health technologies. Note that the term “common drugs” designates health technologies in the CADTH Common Drug Review (CDR), which covers health technologies for conditions such as hypertension, diabetes, and asthma. We considered the above factors because of their potential for health equity implications.

For instance, certain common drugs cover health conditions such as diabetes and hypertension, which disproportionately affect some population groups in Canada (Gagné & Veenstra, 2017). Oncology drugs may require more frequent interactions with health systems for monitoring than some non-oncology drugs (Mercer et al., 2020). Sufficient scientific evidence may not exist on technologies targeting rare diseases, making the patient experience a critical source of evidence in formulating recommendations for these conditions (Conference & May, 2016). Virtually delivered health technologies may not be accessible to those with limited access to digital technologies (Ramel, 2019). Medical devices may raise the concern of access and adjustment to individual needs (Tarricone et al., 2022)

We did not consider HTA reports on digital health technologies. Digital health technologies are different from digital technologies, which we assessed as a modality of patient engagement. Digital health technologies encompass medical devices with built-in digital systems to support various functions in healthcare, including the administration of drugs, diagnostic, monitoring, and predictive testing (Soares et al., 2018; Yao et al., 2022).We excluded them because there is limited patient engagement in HTA about those health technologies(Soares et al., 2018; Tarricone et al., 2022; Yao et al., 2022).

We selected the 2013-2021 timeframe because patient engagement was almost non-existent in Canadian HTA before 2013(Patient Engagement Action Team, 2017). Based on a historical document from the Canadian Agency for Drugs and Health Technologies (CADTH) website, the Common Drug review Panel nominated their first patient representative in 2006 (MacPhail & Shea, 2017). In 2013, a group of patients created the Patient and Community Liaison Forum to promote patient engagement in CADTH's HTA (MacPhail & Shea, 2017). HQO started including patient input in their HTA reports in 2015 (Patient Engagement Action Team, 2017)

5.5.3. Data extraction

We developed a data extraction form with items from the PROGRESS-Plus framework(O'Neill et al., 2014) the checklist to guide equity considerations in HTA (Benkhalti et al., 2021(Benkhalti et al., 2021) and the published literature on characterizing health equity factors in studies (Flanagin et al., 2021; Hosking et al., 2019). We described patient engagement activities with items from the practical guidance for involving stakeholders in health research

(Concannon et al., 2019). A single reviewer extracted data in the included HTA reports; the first author (RS) checked the extracted data for quality control. We provided a detailed description of the variables of interest and data extraction form in **Appendix B.**

5.5.4. Data management and analysis

We used Excel We used Excel for descriptive analysis and the R software package for inferential analysis (Pinheiro, J., Bates, D., & R Core Team., 2022). We utilized Pearson's chi-squared test to determine the degree of associations between patient engagement processes and equity-focused HTA recommendations (CI 95%, p=.05). We used logistic regression to examine the direction and strength of associations between patient engagement processes and equity-focused HTA recommendations. These are dichotomous variables, which take the value of one when the criteria are present and zero otherwise. We expected the coefficient for direct patient engagement or the consensus decision-making model to be greater than zero and statistically significant. Therefore, we will reject the null hypothesis if neither the types of patient engagement nor the decision-making models have a relationship with the likelihood of equity-focused HTA recommendations.

We performed a regression analysis to determine the association between patient engagement processes and equity-focused HTA recommendations (Szumilas, 2010). We did not add a variable for the three different types of reviews. We did not expect the implementation of patient engagement to differ by HTA organization. For example, if HQO or CADTH implemented direct engagement, they would do it similarly. We then calculated the odds ratio (OR) to determine the likelihood of identifying equity-focused recommendations for each type of patient engagement and decision-making model.

5.6. Results

5.6.1. Characteristics of patient engagement processes

We used the practical guidance for involving stakeholders in health research to describe procedural parameters that HTA analysts in CADTH and HQO used to conduct patient engagement. We reviewed the patient input section and patient engagement guidelines from CADTH and HQO websites to characterize patient engagement procedures in the included reports. When conducting the study, CADTH did not require patient organizations to report on their engagement methods to develop patient input. As shown in **Table 4**, we identified the following procedural parameters of patient engagement strategies: types, modes and modalities of engagement, decision-making models, and patients' roles

Types of engagement: We identified two types of patient engagement in the HTA reports: direct and indirect. For direct engagement, HTA analysts collected input directly from individual patients. For indirect engagement, HTA analysts received patient input submissions from patient organizations, who led engagement activities with their members to develop patient input. Patients with experience with the conditions or the health technology provided input. In some cases, healthcare providers provided inputs, which were considered alongside patients' feedback (12%; 7 out of 60). CADTH typically used indirect engagement, while HQO implemented direct engagement to inform their HTA.

Modes of engagement: As mentioned above, an interview was the primary mode of engagement in HQO. As CADTH invited patient organizations to submit feedback on the health technology under consideration, we recorded the mode of engagement for patient organizations. About half of the patient organizations who submitted patient input to CADTH reported on methods of collecting feedback from their members (55%; 22 out of 40). Amongst the patient organizations who wrote their modes of engagement, around 60% used mixed methods to collect feedback from members (59%; 13 out of 22). These organizations used surveys, interviews, and focus groups for patient engagement. The remaining 40% only used surveys to gather patient input from their members (41%; 9 out of 22).

Modalities of engagement: Based on the review of corporate documents, both HTA organizations used digital technologies as the primary modality for engaging patients. Health Quality Ontario (HQO) mainly used telephones to interview patients and caregivers. We also noted that HQO considered in-person engagement in its patient engagement guidelines. Sometimes, HQO uses social media for advertising patient engagement opportunities. CADTH generally posted calls for input on a web portal that allowed patient organizations to submit feedback on health technologies under review.

Although not required to report on their engagement strategies, a third of the patient organizations reported on modality of engagement (31%, 13 out of 40 reports). As shown in **Table 4**, patient organizations used various digital tools to collect input from their members. These included online survey platforms, discussion boards, phones, and social media.

*Models of decision-making, patients' roles, and timing***:** Patients participated in the HTA process as either key informants or members of advisory committees. Key informants are patients who have experience with the health technology under assessment. HTA advisory committees in CADTH and HQO comprise representatives from the HTA institutions, expert panels, and patients with knowledge of HTA. HQO has a representative from the Ministry of Health in its committee. All members serve on their respective HTA committees for a specified term on a volunteer basis. Representatives from the HTA institutions and the Ministry of Health do not participate in decisions about HTA recommendations.

In all the HTA reports, patients contributed to decision-making through an HTA advisory committee representative. However, the ways patients contributed to decision-making differed depending on the types of assessments. In HQO, the patient representative contributed to decision-making through consensus meetings. The advisory committee members used voting when they could not reach a consensus. The patient representative in the Common Drug Review (CDR) committee influenced decision-making through voting. A consensus meeting was held if the vote did not determine a clear-cut decision in adopting a recommendation. The consensus was also the default approach to decision-making in the pan-Canadian Oncology Drug Review (pCODR). As such, a consensus was the most common decision-making model (58%; 35 out of 60 reports) used in formulating HTA recommendations.

Table 4: Summary of patient engagement procedures in HQO, pCODR and CDR.

Types of HTA review	Characteristics	Description	n (%)
Types of patient engagement			
HQO	Direct engagement	HTA organizations directly interviewed individual patients to collect patient input	20 (33%)
CDR and pCODR	Indirect engagement	HTA organizations collected patient input through a submission from patient organizations	40 (67%)
Modes of engagement			
HQO	Interview	HTA analysts conducted interviews with individual patients (20 out of 20)	20 (100%)
CDR and pCODR	Survey	Patient organizations used a questionnaire to collect patient input (9 out of 22)	9 (41%)
CDR and pCODR	Mixed methods	Patient organizations used a combination of 2 or 3 of the following to collect patient input: survey, interview, and focus groups (13 out 22)	13 (55%)
CDR and pCODR	Unknown	Patient organizations did not report methods of collecting patient input (18 out of 40)	18 (45%)
Modalities of engagement			
HQO and CADTH	HTA organizations	HTA organizations use digital technologies for patient engagement activities	60 (100%)
CDR and pCODR	Patient organizations	Number of patient organizations who reported the use of digital technologies (13 out 40)	13 (31%)
CDR and pCODR	Unknown	Number of patient engagement who did not report modalities of engagement (21 out 40)	21 (69%)
Models of decision-making			
HQO and pCODR	Consensus	All members of the HTA advisory committee can contribute to the decision	35 (58%)
CDR	Vote	All members of the HTA advisory committee can vote on the decision	25 (42%)
Roles of patient			
HQO, CDR and pCODR	Key informants	The patient provided their feedback to HTA or patient organizations	60 (100%)
	Members of an advisory committee	The patient contributed to the decision	
Timing of engagement			
HQO, CDR and pCODR	During effectiveness analysis and decision-making	At what stage of the HTA process patient input is elicited	60 (100%)

5.6.2. Characteristics of equity-focused HTA recommendations

We used the items from the PROGRESS-Plus framework to characterize health equity factors reported in patient input and HTA recommendation sections of the included reports. Some of the HQO reports explicitly referred to health equity. The CADTH reports did not have a section on health equity. We did not require reports to refer to health equity to be included in the study. For HTA recommendations, we recorded PROGRESS-Plus items in the rationale and the evidence used to inform the HTA recommendations. This allowed us to categorize a maximum number of HTA reports containing health equity factors.

Our approach to identifying equity-focused recommendations in the HTA reports ensures that we remain inclusive in our coding. For example, if PROGRESS-Plus items were recorded in the HTA recommendations only, less than a third (28%, 17 out of 60 reports) of the included HTA reports would be classified as containing health equity factors compared to (68%; 41 out of 60 reports) when using the procedures explained above. When a PROGRESS-plus item was repeated more than once in either section, we counted this item as one mention to avoid overrepresentation. We identified PROGRESS-Plus items in both patient input (55%; 33 out of 60 reports) and HTA recommendations sections of the reports (68%; 41 out of 60 pieces). HTA and patient organizations have not provided details on how they incorporated equity considerations in the recommendations and patient input.

We identified 12 unique PROGRESS-Plus items across all the included HTA reports, six of which were from the PROGRESS category. These consisted of a place of residence, language, gender, education, socioeconomic status, and social capital. We coded the other six items in the "Plus" category. They consisted of affordability, age, ethical issues, the conditions' severity, treatment logistics, and stigma. We recorded stigma, social capital, and gender in patient input only. We did not find the following items from the PROGRESS framework, race/ethnicity/culture, and religion- in any sections of the included HTA reports.

5.6.3. Description of equity-focused HTA recommendations

We defined equity-focused HTA recommendation as an HTA recommendation that contains at least one PROGRESS-Plus item. As shown in **Table *5***, HQO had the highest proportion of HTA reports with equity-focused HTA recommendations (75%, 15 out of 20 reports). The pan-Canadian Oncology Drug Review (pCODR) (67%, 10 out of 15 reports) and the Common Drug Review (CDR) (64%, 16 out of 25 reports) came in second and third position, respectively.

Table 5: Equity-focused HTA recommendations in the included reports.

Types of HTA review	**Types of patient engagement**	**Models of decision-making**	**N=60**	**Equity-focused HTA recommendations n (%)**
Health Quality Ontario (HQO)	Direct	Consensus	20	15 (75%)
pan-Canadian Oncology Drug Review (pCODR)	Indirect	Consensus	15	10 (67%)
Common Drug Review (CDR)	Indirect	Voting	25	16 (64%)

5.6.4. Health equity factors in patient input and HTA recommendations

We compared the number of PROGRESS-Plus items identified in patient input with those recorded in HTA recommendations. As displayed in **Figure 5** , mentions of PROGRESS-Plus items were more common in the patient input section (total mentions=84; range 1-7) than in the HTA recommendation section of the reports (full mentions= 72; range 1-4). Around two-thirds of the HTA recommendations (68%, 29 out of 41 reports) contained at least one matching PROGRESS-Plus item in patient input.

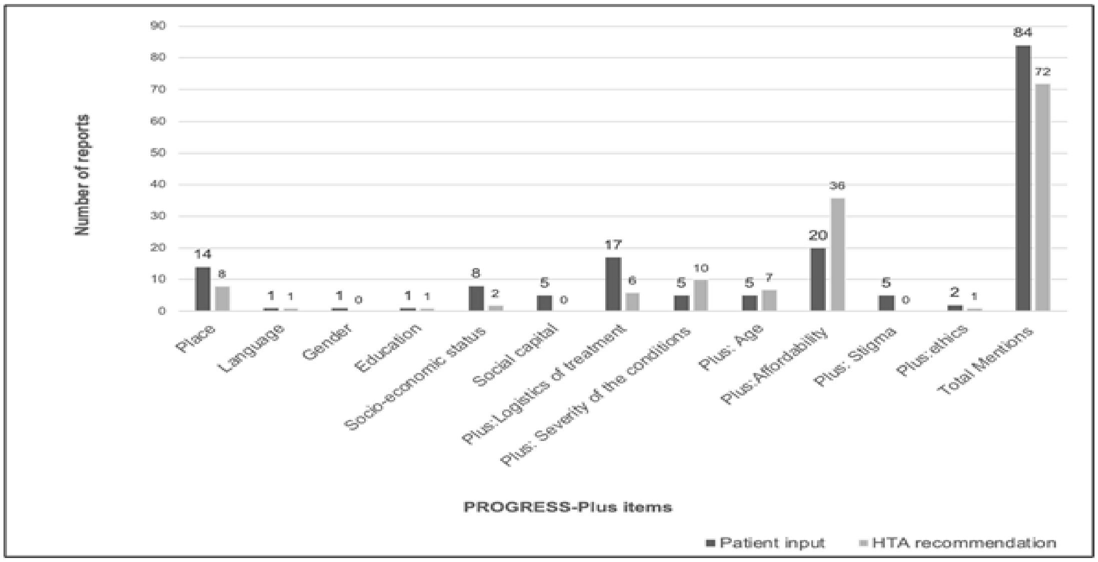

Figure 5: PROGRESS-Plus mentions.

We identified a total of eight PROGRESS-Plus items that are common to both patient input and HTA recommendation sections of the reports. The frequency of the top five of these PROGRESS-Plus items is distributed as follows in patient input and HTA recommendations, respectively: 1) affordability 60% (20 out of 33 reports) compared to 87% (36 out of 41 reports); 2) logistics of treatment 51%, (17 out of 33) compared to logistics of treatment (6 out of 41); 3) place of residence 42 %, (14 out 33 reports) compared to 19% (8out of 41 reports); 4) socioeconomic status 24 % (8 out of 33 reports) compared to 0.5% (2 out of 41 reports); 5) severity of conditions 15% (5 out of 33) compared to the severity of the conditions 24% (10 out 41 reports), 5) age 15% (5 out 33) compared to 17% (7 out of 41).

5.6.5. Association between patient engagement processes and equity-focused HTA recommendations

We used the R package for statistical analysis ((Pinheiro, J., Bates, D., & R Core Team., 2022). As shown in **Table 6**, we found that HTA reviews that used direct patient engagement (OR: 3.85; p-value =.0007; 95% CI [2.40 – 6.20]) and consensus for decision-making (OR: 2.27;

p-value = 0.002; 95% CI [1.35 – 3.84]) were more likely to result in equity-focused HTA recommendations. On the other hand, the likelihood of developing equity-focused HTA recommendations was lower with indirect patient engagement (OR: 0.26; 95% [0.16 – 0.41]) and voting in decision-making (OR: 0.44; 95 % [0.26 – 0.73]), respectively.

More specifically, the likelihood of recording equity-focused HTA recommendations was 2.27 higher when HTA advisory committees used consensus to make HTA decisions than when they used to vote. This scenario was noted in HOQ and pCDOR, with the difference that patient organizations submitted patient input for pCODR reviews. The likelihood of recording equity-focused HTA recommendations in HQO was generally 3.85 higher than the other HTA reports.

Table 6: Inferential statistics

Dependent variable: equity-focused HTA recommendation	**Regression coefficients**	**Odds ratio (OR)**	**Standard error**	**P-values**	**95% CI of odds ratio**
Types of patient engagement					
Direct engagement	1.35	3.85	0.23	0.0007	2.40 – 6.20
Indirect engagement	-1.35	0.26	0.23	0.0007	0.16 – 0.41
Models of decision-making					
Consensus	0.82	2.27	0.26	0.002	1.35 – 3.84
Voting	-0.82	0.44	0.26	0.002	0.26 – 0.73

5.7. Discussion

We used a sample of 60 reports from two HTA Canadian organizations to assess the relationships between patient engagement processes and the inclusion of equity factors in HTA recommendations. We identified two types of patient engagement: direct interviews and indirect engagement through patient group submissions. HTA organizations collected patient input during the effectiveness analysis stage in the HTA process. Patient and HTA organizations engaged patients through digital and in-person modalities, such as document submission and face-to-face meetings. However, patient organizations relied on more diverse digital technologies to engage patients than HTA organizations. Patients contributed to developing recommendations through consensus and voting as members of HTA advisory committees.

We used a broad definition to help capture health equity considerations in the HTA reports. The results suggested that patient engagement played a role in incorporating health equity factors in the included reports. The findings also showed that combining specific patient engagement procedures might increase the identification of health equity factors to inform HTA recommendations. As in previous studies, the results suggested that direct engagement and consensus in decision-making increase the integration of health equity factors in HTA(Bombard et al., 2018b; O'Flaherty et al., 2021; Reynolds et al., 2021).

For example, HTA advisory committees that used consensus as their decision-making model were more likely to consider equity factors in their HTA recommendations. HQO and pCODR were the two reviews with consensus as decision-making models. However, HTA analysts directly interviewed patients to collect input for HQO, whereas patient organizations submitted input for pCODR reviews. The context of the HTA reviews could explain the above results (Bombard et al., 2018b; Lopes et al., 2016). Health jurisdictions -federal versus provincial- might influence the reports of health equity factors in HTA reviews (Lopes et al., 2016; Vreman et al., 2020). HQO's engagement might focus more on addressing implementation issues because of their provincial mandate. In contrast, CADTH HTA recommendations address a national audience and would be further adapted to address contextual problems (Manafo et al., 2018)

Health equity factors in the pCODR reviews, which used consensus for decision-making, could be linked to the history of sustained advocacy around oncologic treatments (Adkins et al., 2017; Korchagina et al., 2019; Mercer et al., 2020). Similarly, a lack of awareness and organized advocacy around certain conditions in the Common Drug Reviews (CDR) pool could explain why PROGRESS-Plus items were less likely to be mentioned in those reports. Common Drug Reviews (CDR) cover conditions such as diabetes, hypertension, mental health, and some rare diseases that are known to affect racialized individuals disproportionately, women, historically stigmatized conditions, and people underrepresented in research(Gagné & Veenstra, 2017; Krieger, 2014; Marmot, 2017; Shimmin et al., 2017).

We noted that the patient input section of the reports contained way more PROGRESS-Plus items than the HTA recommendation section. This observation might reflect a failure to consider patient perspectives reported in input submitted to HTA organizations. Nonetheless, affordability was the top PROGRESS-Plus item in both patient and HTA reports. This is not surprising because the primary outcome of HTA reviews in Canada is to make funding recommendations (Bidonde et al., 2021; Manafo et al., 2018).

Critical items from the PROGRESS framework such as gender, sex, occupation, race/ethnicity/culture, and religion- were not identified amongst the PROGRESS-Plus things recorded in any of the HTA recommendations. Earlier studies showed that gender, culture, access to social capital and discrimination significantly impact health inequities (Bouchard et al., 2015; Krieger, 2014; Marmot, 2017).

5.7.1. Strengths and limitations

We used an explanatory case study approach to elucidate the relationships between patient engagement processes and equity-focused recommendations in two Canadian HTA organizations operating at the federal and provincial levels. The study spotlighted patient engagement as an intervention with distinct processes, which might influence incorporating equity factors in HTA recommendations. We used a systematic process for collecting data with items from validated conceptual frameworks.

Despite these strengths, many limitations are worth considering before utilizing the research findings. The small sample size and the narrowed scope might impede the application of the results to other HTA agencies. The sample size might lead to missing HTA reports with more health equity considerations. We only conduct the study in two agencies in Canada. We cannot know if applicable to other agencies as their contexts are different. However, our hypothesis can be tested in other HTA settings.

We did not add a variable for the three types of HTA review to help increase the power of the analysis. We could not identify a taxonomy of health technologies when doing this research. We did not classify the types of health technologies in each report. If there were a difference due to types of HTA reviews and health technologies, we would not be able to assess it.

Also, a single reviewer extracted the data, and variables were not independent in the analysis. To help reduce errors in data extraction, the first author checked all the data for accuracy and met with data extractors to resolve any discrepancies. Finally, we cannot know how much advisory committee members weigh health equity factors in their final decision.

5.7.2. Implications for practice, policy, and research

The findings elucidate the influence of patient engagement processes in developing equity-focused HTA recommendations and characterize modalities for engaging patient engagement in HTA. The perspectives of patients and HTA analysts on leveraging digital technologies for patient engagement are examined in other articles. HTA and patient organizations can use the findings to inform practice around patient engagement and health equity analysis.

For example, the findings can serve as a foundation for designing patient engagement strategies to increase the integration of health equity factors in HTA. HTA and patient organizations can use those findings to raise awareness in public about the importance of patient engagement in reflecting their input in HTA. Also, patient advocates and the public can use those results to inform their advocacy around increasing their voices in HTA recommendations. Patient advocates can collaborate with HTA organizations on reporting structures for patient input.

The findings have implications for policy-makers who can use them to initiate discussion about expectations of health equity factors in HTA recommendations for their jurisdictions. Future research could investigate the impact of developing equity-focused HTA recommendations on health systems. Additional studies could examine relationships between equity-focused HTA recommendations and funding decisions regarding health technologies.

5.7.3. Conclusion

This is the first study to explore the relationship between patient engagement processes and the development of equity-focused HTA recommendations in Canada. The study reveals that patient engagement that involves HTA analysts directly interviewing patients about health technologies was associated with more significant reporting of equity considerations - PROGRESS-Plus items- in HTA recommendations.

The findings also suggest that HTA organizations need to collaborate more closely with patients to enhance the inclusion of patient perspectives in HTA recommendations. In this vein, the research has paved the way for further research on debunking the complexity of developing equity-focused HTA recommendations in collaboration with patients. Finally, HTA and patient organizations can use those findings to educate the public about opportunities to contribute to designing healthcare systems, which have implications for advancing health equity.

Chapter 6: Manuscript 2

Title: Patients' perspectives on patient engagement to inform equity-focused health technology assessment (HTA) recommendations in the digital era: a grounded theory case study

6.1. Overview of Chapter 6

Chapter 6 is a complete manuscript describing the qualitative study with patients. The manuscript covers the dissertation's second objective, identifying implementation considerations for patient engagement in HTA. Chapter 6 contains the following sections: abstract, background, methods, results, and discussion.

Targeted journal: International Journal of Technology Assessment in Health Care (IJTAHC)

Authors' contributions:

- Rosiane Simeon is the Ph.D. candidate and lead author of the manuscript. Rosiane led the planning, research design, data collection, analysis, and manuscript writing.
- Peter Tugwell and Vivian A. Welch co-supervised the Ph.D. candidate and contributed to the planning, research design and review of the manuscript.
- Ian D. Graham, Kumanan Wilson, Reiner Banken, Shehzad Ali, and Janet H. Roberts are the Thesis Advisory Committee (TAC) members and contributed to the research design and review of the manuscript.
- Anita Rizvi (AR): is a co-author and contributed to data analysis and interpretation of findings.

6.2. Abstract

Background: Health technology assessment (HTA) is a policy analysis approach to develop recommendations for policymakers. Recent research suggests that patient engagement processes can influence the inclusion of equity factors in HTA recommendations. As HTA is context-bound, it is crucial to identify organizational strategies to increase the impact of patient engagement in HTA.

Objective: To identify implementation considerations for patient engagement to inform the development of equity-focused HTA recommendations in two Canadian organizations: the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO).

Methods: We interviewed five patients aged 18 and older who contributed to HTA in CADTH and HQO over the past five years. We combined grounded theory and thematic analysis methods to generate and interpret the findings.

Results: The analysis of interviews revealed patient engagement processes for developing equity-focused HTA recommendations. The research participants highlighted the need for CADTH and HQO to develop strategies to implement more diverse and inclusive patient engagement. To accomplish this, the HTA organizations should consider patients' preferences and capabilities to participate in virtual and in-person engagements.

Conclusion: The findings can inform planning in CADTH, HQO and other organizations with similar characteristics for the rollout of equitable patient engagement in this digital era.

6.3. Introduction

Health technology assessment (HTA) is a policy analysis approach to inform decision-makers on the justification for funding or not a health technology (Angelis et al., 2018; O'Flaherty et al., 2021; O'Rourke et al., 2020). Health technologies include drugs, medical devices, and health services designed to improve health outcomes (O'Rourke et al., 2020; Stevens et al., 2003). HTA organizations, like the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO), have a significant responsibility to build the evidence necessary for informing the decisions of policymakers about health technologies (Manafo et al., 2018). In addition to cost-effectiveness reports, the analysis of patients' lived experiences can help elicit health equity factors to inform the development of HTA recommendations (Benkhalti et al., 2021; Cookson et al., 2017; Culyer & Bombard, 2012). An equity-focused HTA recommendation highlights the need to address social factors that impede the effectiveness of health technologies in improving health outcomes (Benkhalti et al., 2021; Braveman & Gruskin, 2003; Tugwell et al., 2006b).

Patient engagement in HTA typically involves developing partnerships between HTA institutions and individual patients or patient groups (Facey Helle et al., 2017; Manafo et al., 2018; McCoy et al., 2019). HTA analysts can engage patients at any stage in studying health technologies' attributes, including determining the scope of HTA, assessing evidence and formulating recommendations (Facey Helle et al., 2017; Wahlster et al., 2017). Patient engagement strategies vary with the context of HTA organizations and the characteristics of the health technology (Cavaller-Bellaubi et al., 2021; Facey Helle et al., 2017; Wahlster et al., 2017).

For example, the two Canadian organizations that are the subject of this case study use different mechanisms to engage patients in their HTA processes (Conference & May 2016;

Manafo et al., 2018). HQO collects patient inputs through key informant interviews, and patients contribute to decision-making by participating on committees. The committees must reach a consensus on decisions regarding HTA recommendations (Manafo et al., 2018). CADTH uses patient group submission for some of its HTA reviews; patients also contribute to decision-making through votes and consensus depending on the types of HTA reviews (Conference & May 2016; Manafo et al., 2018).

Studies discuss the need for improving inclusion in patient engagement processes (Facey, 2019; Mercer et al., 2020; Reynolds et al., 2021). Research suggests that inclusive patient engagement processes help acknowledge differences in knowledge, skills and interests among patients and researchers (Facey, 2019; Mercer et al., 2020; Reynolds et al., 2021). Acknowledging these differences can lead to diverse perspectives and new understandings surrounding health outcome (Charlier et al., 2017; Reynolds et al., 2021; Roche et al., 2020). Inclusive patient engagement processes can eventually lead to integrating more health equity factors in HTA recommendations (Charlier et al., 2017; Reynolds et al., 2021; Roche et al., 2020). Inclusive patient engagement consists of creating a research environment where research teams manage power dynamics and research processes in ways that enable patients to participate positively and productively (Black et al., 2018; Cavaller-Bellaubi et al., 2021; Khuntia et al., 2022; Reynolds et al., 2021; Shimmin et al., 2017).

Another aspect of inclusion in research is demonstrating to patients that their input is valued (Biddle et al., 2021; Cavaller-Bellaubi et al., 2021; de Wit et al., 2020; Snow et al., 2018). For example, two recent studies found that allocating a budget to patient engagement activities and providing travel accommodations helped demonstrate to patients the value of their input in research (Biddle et al., 2021; Cavaller-Bellaubi et al., 2021). In addition to these enabling factors, other studies reported that patients felt that researchers valued their contribution when considering their input while maintaining rigour in the research process (Black et al., 2018; Chudyk et al., 2022; Gill et al., 2018; Gunn et al., 2021).

Inclusive patient engagement can help integrate patient concerns and opinions into knowledge products such as HTA recommendations (Cavaller-Bellaubi et al., 2021; O'Flaherty et al., 2021). Researchers and patient advocates continue to raise concerns about how HTA organizations use patient input in HTA (de Wit et al., 2020; Reynolds et al., 2021). Also, there is little research on strategies for recruiting and supporting patients to inform the development of equity-focused HTA recommendations (Facey, 2019; Reinius et al., 2022).

Digital technologies can enable broader citizen engagement and bring diverse perspectives into mainstream debates about health technologies (Kent & Yellowlees, 2015; Mazanderani et al., 2013; Mitchell & Kan, 2019). It is, however, essential to consider the digital divide, which may arise from differences in access, availability, and ability to manipulate digital technologies (Lai & Widmar, 2020). Digital technologies cover various information and communication technologies to support data production, management, and processing (Aziz & Madani, 2015; P. Sharma & Kaur, 2017). Digital technologies are helping patients to openly share their experiences with health technologies (Mazanderani et al., 2013; Mitchell & Kan, 2019). HTA organizations should explore the unprecedented opportunities digital technologies offer to engage patients from diverse backgrounds and need to inform HTA recommendations (Aziz & Madani, 2015; Brewer et al., 2020; Mitchell & Kan, 2019).

6.4. Objective and research questions

This qualitative case study aims to identify implementation considerations for patient engagement to inform the development of equity-focused HTA recommendations. This case study addressed the following research questions:

1. How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?
2. How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?

6.5. Research design and methods

6.5.1. Research design and rationale

The research design consists of an explanatory case study of patient engagement processes for HTA in two Canadian organizations: CADTH and HQO. We selected CADTH and HQO as a case because they are operating at the federal and provincial government levels, respectively. The study will help highlight the particularity of implementing patient engagement for HTA in the Canadian context. The explanatory case study approach can help explain patient engagement's role in developing equity-focused HTA recommendations in CADTH and HQO (Crowe et al., 2011b; Fisher & Ziviani, 2004). Moreover, it promotes collaboration between researchers and research participants (Crowe et al., 2011b; Fisher & Ziviani, 2004), facilitating the use of research findings for organizational change (Graham et al., 2006).

We used a pragmatic worldview to guide the qualitative research. Pragmatic philosophy to research helps frame the production of knowledge from participants' perspectives and focuses on applying the research results to tackle a specific problem (Creswell & Clark, 2017; Kaushik & Walsh, 2019). It also acknowledges that truth is context-bound and varies with one’s perspective, a constructivist paradigm that compasses the social construction of reality (Creswell & Clark, 2017; Kaushik & Walsh, 2019).

6.5.2. Researchers' positionality statement

We chose a pragmatic approach to designing the study because it enables participants to share their perspectives and impact research narratives (Creswell & Clark, 2007; Kaushik & Walsh, 2019). Additionally, the pragmatic worldview is well-aligned with the focus of the population health field, which has populations directly affected by health interventions at the center of research for developing policy alternatives (Carpiano & Daley, 2006; Kaushik & Walsh, 2019). Finally, we believed that using a pragmatic approach in this study could help acknowledge the diversity of needs and interests amongst patients, HTA analysts, researchers, and policy-makers, who are critical in improving the development of equity-focused HTA recommendations.

6.5.3. Researchers' characteristics and reflexivity

The research The research team comprises experienced academics and researchers knowledgeable about the Canadian HTA context. Before leading this research, the principal investigator (RS) practiced family medicine in Haiti and obtained a Master of Public Health in the United States of America. Over the past nine years, she has worked as a researcher and policy analyst in Canada. All these experiences have moulded her research interest and analytical skills (Dodgson, 2019).

6.5.4. Context of the study

The qualitative explanatory case study is the second paper of a publication series to explore relationships between patient engagement processes and the development of equity-focused HTA recommendations. The series also identifies implementation considerations for patient engagement in HTA. We use an explanatory mixed methods research design containing one cross-sectional quantitative study and one qualitative study. In the first paper, we describe the findings of the quantitative research. We report the qualitative study results in the second and third papers, focusing on the perspectives of patients and HTA analysts, respectively.

The first paper identifies critical elements of patient engagement processes in CADTH and HQO. We also examine the association between these processes and the development of equity-focused HTA recommendations. The first study's findings reveal that patient engagement processes may have influenced the integration and reporting of health equity factors in HTA recommendations. HTA reviews where HTA analysts directly interview patients and use consensus as decision-making had more equity-focused HTA recommendations. Also, HTA reviews that use patient group submissions and consensus for decision-making were more likely to have equity-focused recommendations than HTA reviews that use the same types of patient engagement but employ vote for decision-making instead of consensus.

In this second paper, we describe patients' perspectives on implementation considerations around patient engagement strategies to inform the development of equity-focused HTA recommendations. In the third paper, we discuss additional findings of the qualitative study relating to the perspectives of HTA analysts.

6.5.5. Ethics approval

RS obtained ethical approval from the University of Ottawa to implement the study protocol; certificate of ethics approval number: H-07-20-5948.

6.5.6. Sampling strategy

RS used a snowball method to identify five English-speaking patients who had contributed to HTA processes in CADTH and HQO over the past five years. The sample size was mainly based on feasibility because we focused on only two HTA organizations. We recognized that this project might not be a priority for eligible participants as they must deal with their health issues (Aluwihare-Samaranayake, 2012; Stephanie A Kraft et al., 2021). For example, to show respect for the participants' time, we stopped contacting them if they did not reply to the first reminder email ((Aluwihare-Samaranayake, 2012). We also recruited research participants based on emerging ideas from the first two interviews (Chun Tie et al., 2019). Some of the participants have experience with CADTH and HQO. We reached saturation after five interviews as some participants helped identify issues and solutions pertinent to both organizations.

6.5.7. Interview guides, data collection, processing, and management

RS used findings from the quantitative study to develop an interview guide to conduct semi-structured interviews between February 2022 and June 2022 through Microsoft Teams and Zoom. All participants provided either written or verbal consent before being interviewed. The consent form and interview guide may be found in **Appendix C** and **Appendix D**

Once RS had completed an interview, she uploaded the recording to Otter.ai, a software for verbatim transcription, to transcribe the interview audio file (Otter.ai, 2020). RS reviewed the interview transcripts for accuracy and relabeled interview excerpts to preserve confidentiality and protect the research participants' privacy. After this step, RS uploaded the transcript into NVivo 12 Plus (Castleberry, 2014) to organize, identify, analyze and describe themes from the data set (Bradley et al., 2007; Nowell et al., 2017). RS used the Standards for Reporting Qualitative Research (SRQR) to develop the study report (O'Brien et al., 2014).

6.5.8. Data analysis

RS combined grounded theory and thematic analysis methods to analyze and collect interview data, similar to the study by (Wilkinson-Stokes et al., 2023). Combining grounded theory with a thematic analysis approach provides more flexibility for analyzing and interpreting the data with an applied focus (Nowell et al., 2017; Wilkinson-Stokes et al., 2023). Grounded theory methods help generate possible explanations –a theory- of a phenomenon using concurrent data generation, collection, and analysis through various stages of coding (Bradley et al., 2007; Chun Tie et al., 2019). It is an iterative process involving inductive reasoning to identify themes in the data and modify the interview guide (Chun Tie et al., 2019). It can also guide the identification of new interviewees to elucidate ideas and concepts that emerged while coding interview data (Chun Tie et al., 2019; Creswell & Clark, 2017). This sampling strategy is called theoretical sampling (Bradley et al., 2007; Chun Tie et al., 2019).

Unlike grounded theory, thematic analysis does not aim to identify a theory (Nowell et al., 2017). The thematic analysis allows the examination of research participants' perspectives to generate insights using either inductive or deductive coding in a structured manner (Bradley et al., 2007; Nowell et al., 2017; Wilkinson-Stokes et al., 2023). In this way, the two methods overlap in that thematic analysis helps identify, categorize, and describe the themes connected to developing the grounded theory (Chun Tie et al., 2019; Nowell et al., 2017). With the thematic analysis, researchers can use deductive coding to further categorize themes based on existing conceptual frameworks (Nowell et al., 2017; Wilkinson-Stokes et al., 2023). This variable-oriented approach can help demonstrate the transferability of the findings (Maher et al., 2018; Nowell et al., 2017; Wilkinson-Stokes et al., 2023).

For this study, RS used grounded theory separately and concurrently with thematic analysis to identify themes and determine their relationships. RS conducted the data analysis in two steps: 1) develop thematic categories with grounded theory and 2) organize thematic categories with a conceptual framework

1) Develop thematic categories using the grounded theory approach

RS used the techniques described by Bradley and colleagues to analyze interview data and develop a thematic framework, which consists of identifying and organizing themes to determine their relationships (Bradley et al., 2007; Chun Tie et al., 2019; Nowell et al., 2017)). RS transferred and analyzed the interviews line-by-line in NVivo and then used iterative analysis to adapt the interview guides. This inductive process allowed seamless connections between several versions of the interview guide, regularly adjusted to help either clarify previously identified themes or develop new ones. RS used memos as informal analytic notes to help keep track of connections between interview data and theoretical thematic categories.

2) Organize thematic categories with a conceptual framework

RS used a conceptual framework composed of items from the knowledge-to-action (KTA) cycle and the technology acceptance model (TAM) (Graham et al., 2006; Taherdoost, 2018). We describe the application of the KTA and TAM to the study in the paragraphs below. As illustrated in **Figure 4,** we used the TAM to supplement the KTA for describing barriers and facilitators to implement patient engagement processes to inform the equity-focused HTA recommendations. The deductive approach can draw attention to applying evidence-based practices to use the research findings.

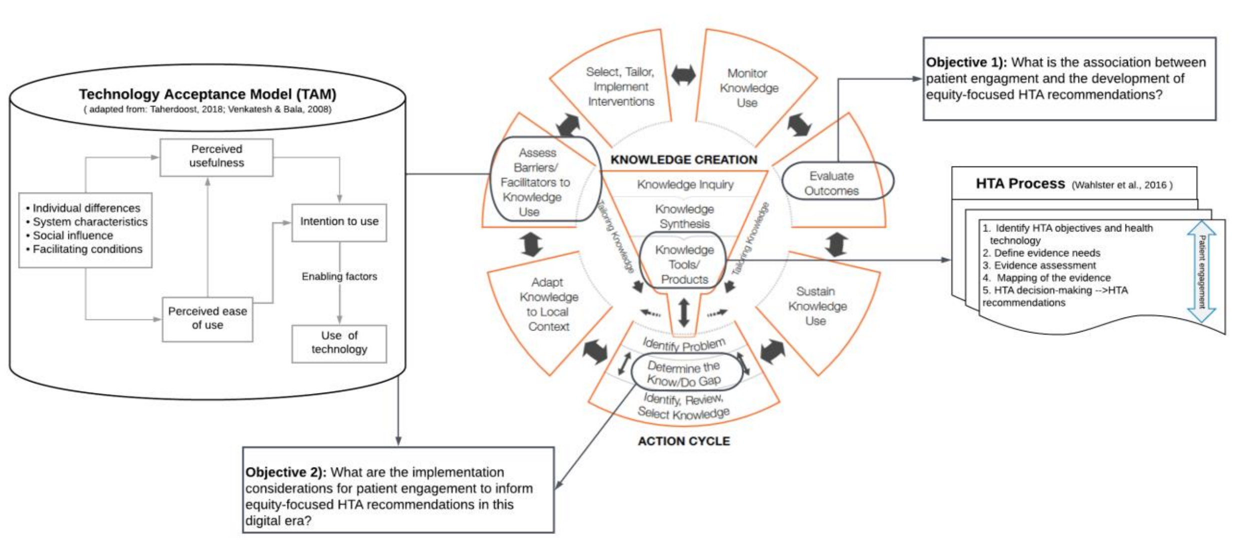

Figure 4: Application of KTA cycle to the thesis (Adapted from Graham et al. 2006).

Determine the Know/Do gap: In this phase, researchers and stakeholders work together to identify a practice problem, review evidence related to tackling this problem and determine what needs to be done to put that knowledge into practice to address the previously identified shortcomings (Graham et al., 2006). In this qualitative study, *the know/do gap* consists of considerations for implementing patient engagement strategies to inform the integration of health equity factors in HTA recommendations.

b) *Assess barriers and facilitators to using knowledge:* This phase describes the various factors that may impact the effective use of expertise to achieve the intended changes (Graham et al., 2006). We used a behavioural model, the Technology Acceptance Model (TAM), to help validate the concepts identified and strengthen the transferability of knowledge generated from the data analysis (Graham et al., 2006; Taherdoost, 2018). The TAM provides a theoretical framework to explain the adoption of new technologies. It suggests that using a specific technology depends on multiple parameters, including the characteristics of the technology, social influence, facilitating conditions and the characteristics of the intended users (Taherdoost, 2018; Venkatesh & Bala, 2008). These parameters affect users' perceptions of the technology's usefulness and ease (Taherdoost, 2018).

Perceived usefulness is related to the functionality of the technology in helping to solve the identified problem, whereas perceived ease of use concerns efforts required to effectively address the issue at hand (Taherdoost, 2018). Based on the TAM, perceived usefulness and ease determine the intention to use or accept a specific technology. Intention to use coupled with enabling conditions such as organizational support will increase the chance of people using the technology (Taherdoost, 2018).

6.5.9. Trustworthiness

We used the strategies described by Maher and colleagues to ensure rigour in the research process by enhancing its transferability, dependability, credibility, and confirmability. For transferability, we used purposeful sampling to recruit participants with experience in HQO and CADTH. We also detailed the study's context, participants' characteristics, data collection process, and analysis. We used a conceptual framework to demonstrate the applicability of the findings to implement organizational change Our approach will ensure that knowledge users have enough details about the study to assess whether the findings are transferable to their context.

We shared the philosophical approach guiding the research and discussed how the research team's skills, training, and work experiences could influence the dependability of the research. To uphold the credibility of the findings generated from this study, we did member-checking to validate the results with participants and had another researcher review the coding. Finally, we used feedback from the thesis advisory committee members to improve the analysis.

Member-checking also contribute to ensuring confirmability of the findings (Bradley et al., 2007; Malterud et al., 2016). RS organized all the data into a thematic category and sent interviewees a summary of themes for feedback on interpreting their responses. RS applied the process described above to coding the data. AR checked the different versions of the analysis to ensure rigour and consistency in the research and interpretation of the data. For confirmability, we described the data management process and analytical approach. We also provided quotes to support our analysis.

6.6. Findings

6.6.1. Characteristics of research participants

RS identified potential research participants on the websites of CADTH and HQO. RS approached a total of seven potential interviewees throughout the data collection phase. Two of them did not respond. RS interviewed five patients for this study: two were members of a patient advocacy group, three had experience with CADTH and HQO HTA processes, and two had only experience with the HQO process. All participants signed a consent form to participate in the study. For privacy protection, the ethics officer advised against asking participants about their race, age, sex, gender, or health conditions. However, some of these demographic characteristics were revealed later in our conversations about patient engagement in HTA. Also, because the Canadian HTA community is small, we did not report on research participants' roles in protecting their identity.

6.6.2. Synthesis and interpretation

In this section, In this section, we described the perspectives of five patients who had participated in patient engagement to inform HTA recommendations in CADTH and HQO. As indicated earlier, we organized the findings using selected components of the knowledge-to-action (KTA) cycle and the technology acceptance model (TAM). First, we described the research participants' perceptions about the role of patient engagement in developing equity-focused HTA recommendations. Then, we discussed research participants' experiences and expectations about patient engagement in HTA. Finally, we discussed critical considerations for implementing patient engagement strategies to inform the development of equity-focused HTA recommendations in this digital era. **Table 7** presents an overview of analytical categories and their matching research questions. To manage the length of the manuscript, we used only one quote to illustrate each topic under a subtheme. Additional illustrative quotes are available in **Appendix E**.

Table 7: Overview of analytical categories by research question.

Themes	Sub-themes
Q1: How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?	
1. Perceptions about the role of patient engagement in the development of equity-focused HTA recommendations	• Importance of patient engagement to the HTA process • Patient engagement as the unique source of evidence • Meaning /implications of health equity • Limitations of current health equity analysis in HTA (Challenges)
2. Perceptions about diverse and inclusive patient engagement strategies in HTA	• Expectations about varied and inclusive patient engagement strategies • Positive experiences with current engagement processes/ inclusive engagement
3. Build trust in the HTA process	• Increase cultural competencies in HTA • Provide practical support to patients
Q2: How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?	
4. Critical considerations for implementing digitally enabled patient engagement strategies for equity-focused HTA recommendations.	• Characteristics of digital technologies for inclusion of people with different abilities • Advantages of digitally-enabled engagement (Perceived usefulness and ease of use) • Advantages of in-person meetings • Practical and ethical issues in using digital technologies (Digital divide) • Mixed modalities for diverse and inclusive engagement in HTA (Recommendations)

Theme 1: Patient engagement processes in health technology assessment (HTA)

In this section, we described the interviewees' perceptions about the importance of patient engagement to inform equity-focused HTA recommendations and recommendations for increasing the impact of HTA recommendations on health equity.

Subtheme 1: Importance of patient engagement in the HTA process

Some interviewees explained that patient engagement represents a means for patients and caregivers to characterize essential health outcomes, advocate for access to health technologies and share experiences with health systems in general. The following research participant explained: "*Patient engagement is extremely key because people want to get out of their system all the trauma they have, and even the good things that happened to them within the healthcare system." [Patient 02]*

One interviewee talked about how their lived experiences represent a unique source of evidence for researchers. The interviewee argues that their experiences handling their illness and role as caregivers navigating healthcare systems allowed patients to acquire invaluable knowledge about health conditions and technologies. Below, they explained that patients could help researchers better appreciate the complexity of living with a health condition and all the intricacies involved in effectively using health technologies: *"Medical doctors must learn everything from the common cold to pancreatic cancer. I had to learn one thing, autism. Let's say, whatever your patient is; they must learn one thing. I said I would challenge you. Despite all your education, I know more about my one thing than you do." [Patient 05]*

Another interviewee recounted how patient engagement helped highlight details about the day-to-day difficulties involved in accessing and using health technologies, including transportation and conservation of drugs. This interviewee was also quick to draw attention to the difference between their knowledge and that of healthcare providers to highlight the importance of gathering patient input to inform HTA recommendations: "*For example, I've had people say: look, I'm a physician, I know my patients, I understand their needs. And my response to that often looks like this; I've got a 15-minute appointment that, I've driven from Ottawa to Toronto, and there are only certain things I will discuss with you, and maybe I'm not mentioning that travelling with the refrigerated drug is problematic and just reminding them that my interactions are minimal and that my voice can't always come through in those appointments." [Patient 01]*

Another research participant suggested that it might be more practical to collect feedback on the effectiveness of health technologies from patients instead of relying on healthcare professionals to speak on behalf of their patients. Patients do not necessarily trust healthcare providers as credible voices to raise their concerns to policymakers. The excerpt below elucidates patients' perspective about mistrust in healthcare providers:*" The parents felt the nurse was mishandling the child and raised that concern, but the team of experts was working on covering this up. And I don't think it's an isolated case. Because they seem to cover things up a lot, especially when it has to do with babies of colour." [Patient 04]*

Subtheme 2: Meaning of equity-focused HTA recommendations

As we reported above, the interviewees described patient engagement as a critical source of evidence to inform HTA recommendations. However, some research participants said it is essential to clarify the meaning of health equity to help HTA researchers better integrate their concerns into HTA recommendations. For example, the interviewee below doubted that current health equity analysis could help incorporate their day-to-day problems in HTA recommendations. They said the scope of health equity analysis is too focused on socioeconomic factors such as race, income, and location: *"It is important to understand what health equity means. What does vulnerability mean? For some people, it is mainly about our racial issues. Racial issues are fundamental, but what about those who don't know how to advocate for another choice, education? Could it be a cultural issue too? The doctor knows best; we are in a paternalistic system. It could be the ability to Google something?" [Patient 01]*

The following interviewee expressed the importance of looking at factors such as race-based stigma and communications with healthcare providers to inform health equity analysis: *"When you look at health equity, you need to know that people of colour experience a lot of racism in the healthcare systems, and they have poor health outcomes; because doctors and nurses do not always understand how we [people of colour] communicate."* [*Patient 04]*

Another interviewee explained that multiple jurisdictions would have to work together to develop synergistic strategies to ensure that health technologies contribute effectively to improving health equity. The interviewee clarified the implications of developing equity-focused HTA recommendations: *"I also think that health equity is something that you can't apply to one field only. If equity isn't applied across every interconnected field, it's like a chair that should have four legs having only two; it needs the other two to stand up strongly. So, it's necessary for HTA, but only in conjunction with other equity measures". [Patient 02]*

Theme 2: Perceptions about inclusive patient engagement strategies

Under this theme, we described research participants' perceptions and experiences regarding diversity and inclusion in patient engagement. We also discussed interviewees' recommendations to HTA organizations about patient engagement to inform health equity analysis in HTA. The interviewees discussed the need to build trust in the HTA process and provide practical support, such as travel accommodations and compensation for patients participating in HTA.

Subtheme 1: Meaning of diversity in patient engagement

The research participants discussed the need for HTA and patient organizations to support patients from diverse backgrounds to contribute to the HTA process. They depicted diversity in patient engagement as a process that enables individuals from all walks of life to tell their stories about their health conditions and lived experiences with health technologies. One interviewee explained: "*They [HTA organization] were asking me to come on board and give my opinion for certain health issues. I asked them to reach out and include more First Nations voices. After saying this, they stop reaching out to me. So, it can be an uphill struggle to get involved if you're the kind of person who wants to center equity." [Patient 02]*

In addition to the importance of including diverse people impacted by health technologies, another interviewee said HTA organizations should also consider the variety of needs stemming from living with a health condition in their engagement activities. This interviewee explained the importance of diversity in patient engagement in the following way: "*A big part [of patient engagement] for me is understanding the patient's vulnerability, regardless of social background." [Patient 01]*

Some research participants explained that individual patients and patient organizations could also contribute to increasing diversity in patient engagement. The interviewee below recounted their commitment to encouraging their peers to participate in patient engagement: *"I'm hoping to build a community where people know that they can come for support. People are so ashamed of having COPD because it's from smoking." [Patient 03]*

Subtheme 2: Inclusive and positive patient engagement experiences

In general, the interviewees said they had a positive experience in patient engagement. The research participant below said their collaboration with HTA analysts was respectful: *"Look, we had disagreements and asked many important questions, but I've never sat at a meeting where I felt that somebody was trying to disregard me." [Patient 01]*

Another interviewee said they felt patient engagement is valued when researchers provide feedback on their input and see that input reflected in recommendations to policymakers. The interviewee recounted their experience: "*You knew from the interactions, from the questions being asked, that you were giving them an insight into something they've never thought about. We know what we did as a group directly impacted material that went out, in things that would show up at the hospital level regarding the quality plans because they flow down from the HTA organization." [Patient 05]*

Subtheme 3: Build trust in the HTA process

a) Increase cultural competencies in staff

Research participants pointed to a lack of diversity amongst HTA analysts as a challenge for inclusive patient engagement. Another research participant suggested that HTA organizations should hire HTA analysts with diverse backgrounds and abilities to reflect the diversity of patients impacted by HTA recommendations. They explained that HTA analysts with diverse backgrounds could help increase trust in the HTA process: "*One thing that a lot of people in the disabled community will say is that people don't realize that they're just not disabled yet. Governments and people in power must be receptive to others entering this healthcare system." [Patient 02]*

The following interviewee expressed the need for increasing cultural competencies amongst HTA analysts. They said this could help implement more culturally sensitive patient engagement: "*I don't think they consider cultural competencies in HTA. The people I talked to are good, but honestly, they don't seem to understand how I communicate the cultural aspects of healthcare experiences." [Patient 04]*

b) Provide practical support to patients

Some interviewees recommended HTA organizations allocate more funding to patient engagement activities. They said more funding could help increase the diversity of people contributing to HTA: *"This HTA organization is quite interesting, but it had no budget to do things in the scope." [Patient 05]*

Theme 4: Considerations for digitally-enabled patient engagement

As a reminder, we used the TAM to characterize barriers and facilitators to using digital technologies for diverse and inclusive patient engagement. The TAM suggests that the usefulness and ease of use influence the intention to use technology; and that certain contextual factors can interfere with adopting technology. We defined digital technologies as all electronic technologies (e-technologies) and applications that use information in numeric code, including telephone, mobile devices and the internet (Aziz & Madani, 2015). We discussed digital technologies' key characteristics and functions in supporting interactive and non-interactive communications through the internet and the web, which sustain the networking of computers and individuals (Aziz & Madani, 2015; P. Sharma & Kaur, 2017). Below we discussed research participants' perceptions of using digital technologies for patient engagement to inform the development of equity-focused HTA recommendations.

Subtheme 1: Characteristics of digital technologies

Research participants highlighted how some features of digital technologies could enable patients from diverse backgrounds and needs to share their lived experiences with health technologies. Overall, the study participants believed that digital technologies could help increase the reach of patient engagement to allow more diverse patients to contribute to the HTA. They also thought that digital technologies could enhance interaction and interconnectedness between patients and HTA organizations.

The interviewees also appreciated the flexibility of digital technologies when providing immediate accommodations for people who might experience communication challenges during patient engagement sessions. We regrouped research participants' perspectives about digital technologies' characteristics into two categories: perceived usefulness and perceived ease of use.

a) Perceived usefulness

Increase outreach: The interviewees said digital technologies could help identify, recruit, and promote active participation of patients with diverse backgrounds and needs in the HTA process. They said digital technologies could facilitate peer support and connection within patient organizations, especially for patients whose fragile health might require social distancing for increased protection: *"Social media could be an amazing way to connect with other differently-abled people. Especially with Zoom taking off in the last couple of years, a lot of people who are disabled have found that accessibility has grown". [Patient 02]*

Another research participant noted that digital technologies could help establish emotional connections between patients and members of HTA committees: *"Virtual meetings have opened the door to a bigger diversity of voices. So instead of reading a report, the advisory committee could hear the voices of people with those conditions. That would be very powerful." [Patient 01]*

b) Perceived ease of use

Research participants said that built-in interactive functions in digital technologies could help reduce barriers for people with different abilities. Some patients might be able to participate in engagement sessions because online setups offer more flexibility and adaptability in terms of immediate accommodation than in-person meetings. An interviewee explained: "*For somebody with selective mutism, they have a lot of anxiety; let them be present as they can. They don't have to turn on their camera; they can just sign in. And often this is very helped by digital technologies." [Patient 02]*

Subtheme 2: Enabling factors to use digital technologies for patient engagement

As discussed in the previous paragraphs, the interviewees recognized the potential of digital technologies to help increase the diversity of people from recruitment to participate in patient engagement. However, they offered some caveats and suggested solutions for using digital technologies to implement equitable patient engagement. They discussed practical issues such as the availability and accessibility of digital technologies, which can eventually pose ethical issues if ignored when deploying digitally enabled patient engagement. The research participants also raised ethical issues related to ensuring a safe space for conducting a patient meeting in a virtual environment.

a) Level of comfort with digital technologies

The interviewees explained that some people might opt out of virtual meetings because they are uncomfortable using digital technologies. As an interviewee suggested, HTA organizations could guide patients on manipulating digital technologies in their engagement sessions. Providing support in using digital technologies could help patients feel more comfortable participating in virtual engagement: *"Overall, many people are afraid of technology* *and won't try it. So that eliminates some of the people who would benefit from it. And so, in that way, a little training would go a long way." [Patient 03]*

b) Access to digital technologies

Some patients may live in places with limited access to the Internet. As exemplified below, this interviewee felt that their input mattered to the HTA process when HTA provided travel accommodations that enabled them to share their experiences: *"They flew people down from the North, from First Nations reservations. And so, one person said, If I weren't flown down here, I would not have the opportunity to tell my story. The Internet is very bad where I live." [Patient 02]*

c) Preferences for in-person meetings

A research participant highlighted the value of in-person meetings for promoting productive patient engagement: *"Consensus building table is better in-person. There's no doubt about it. It's a lot of body language, facial expressions, and maybe a side conversation during the break. I think it's tougher to read the virtual room for project planning and consensus building." [Patient 01]*

d) Safe space for patient engagement

Another interviewee highlighted the importance of creating a safe space for patients to share their stories, whether online or in-person engagement: *"We need to make a concerted effort to enable people to tell their stories, to see each other from a more diverse perspective, instead of just black and white. Digital technology just emphasizes the human experience." [Patient 02]*

6.6. Discussion

6.6.1. Summary of findings

We used a grounded theory approach to analyze interviews of five patients to elucidate considerations for implementing patient engagement that can inform the development of equity-focused HTA recommendations. All the research participants had previously provided patient input to inform HTA recommendations in CADTH and HQO. The research participants have discussed the need for HTA organizations to factor in the following implementation considerations in their patient engagement strategies: a) the role of patient engagement in HTA; b) the meaning of health equity in HTA; c) diverse and inclusive engagement; d) digital and non-digital modalities of engagement.

Role of patient engagement in HTA: The research participants have discussed the importance of patient engagement in HTA. They said their lived experience could reveal truly unique perspectives about health technologies; because of their learning in living and navigating healthcare systems with their health conditions. Other studies reported similar findings, which demonstrated that patient input was key in characterizing the burden of health conditions and identifying important health outcomes from using health technologies (Gill et al., 2018; Gunn et al., 2021; Reynolds et al., 2021).

Meaning of health equity in HTA: The interviewees also explained that the scope of health equity analysis in HTA does not necessarily capture their concerns. Some research participants thought that health equity analysis should focus more on eliciting barriers to using health technologies, such as cultural issues, education, gender, logistics and race. Studies showed that such concerns could negate the positive impact of health technologies on advancing health equity (Culyer, 2016; Hoch et al., 2021; Mazor et al., 2016; Yao et al., 2022).

Diverse and inclusive engagement: The research participants also discussed their perceptions of diversity and inclusion in HTA. They explained that HTA organizations should engage various people impacted by HTA recommendations. The research participants also described inclusion in patient engagement as a commitment of HTA organizations to create positive experiences, elicit patients' needs, and demonstrate the value of engaging patients in their processes. For those reasons, the interviewees advised HTA organizations to promote trust in their HTA and provide practical support to patients to enable them to contribute to HTA. For example, the interviewees explained that HTA organizations could hire HTA analysts with diverse backgrounds and abilities and compensate some patients for encouraging them to participate in HTA.

Digital and non-digital modalities for engagement: The interviewees advised HTA organizations to use digital technologies with in-person arrangements to minimize the impact of the digital divide. The interviewees explained digital technologies could help HTA organizations improve engagement procedures such as identification, recruitment, and collaboration. Other digital technologies' features, such as connectedness, flexibility, and adaptability, could attract more patients and accommodate people with different communication needs.

Nevertheless, the research participants warned against solely relying on digital technologies to ensure equitable access to patient engagement. Some patients may not have access to digital technologies because of where they live, their disabilities or their income. Others may not be comfortable manipulating digital technologies. Therefore, HTA organizations should offer in-person options in their engagement strategies. In alignment with considering the digital divide, HTA organizations should provide travel accommodations to enable the participation of patients who might need this practical support to be able to contribute to HTA.

6.6.2. Strengths and limitations

A critical strength of this study is that it addresses specific Know/Do gaps in patient engagement processes at CADTH and HQO. This strength also introduces the limitation of preventing the generalization of the findings. The scope of the study is very narrow, and the results may not be transferable to non-Canadian HTA institutions. Additionally, the context and jurisdiction of CADTH and HQO are different- federal versus provincial mandates-; thus, each organization might need to assess how to implement digitally-enabled patient engagement that could lead to developing HTA recommendations. The findings of this case study can inform this assessment.

There are additional limitations that are inherent to the research design and methods. By design, only patients who contributed to HTA in CADTH and HQO were eligible for this study. This criterion made the pool of participants we could recruit very small. Only one researcher analyzed the interviews. To minimize the impact of those limitations, we integrated different theoretical perspectives into our analysis (Nelson, 2017). We also did member-checking to validate the interpretation of the interview data (Dodgson, 2019; Malterud et al., 2016).

6.6.3. Implications for practice and research

The findings have implications for practice and research. They can inform strategies around building capacity for inclusive patient engagement. They can also help leverage digital technologies to sustain patient engagement that can support the development of HTA recommendations. However, the results warrant further investigation to clarify the scope of health equity analysis n HTA. Earlier studies identified the need for implementing inclusive patient engagement to influence health research products like HTA recommendations(Facey, 2019; Reynolds et al., 2021; Shimmin et al., 2017). Some studies suggested that applying health equity and a social justice lens to patient engagement can help demonstrate the value of patient engagement and inform equity-focused health policies (Black et al., 2018; Davis et al., 2019; Roche et al., 2020; Snow et al., 2018). We discussed the implications of the findings in the following paragraphs. We summarized the study's key findings and their implications for practice in **Table 8**.

Table 8: Summary of key findings and their implications for practice.

Q1: How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?	Q2: How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?
HTA organizations should build organizational to sustain inclusive patient engagement	HTA organizations should use digital technologies in multi-pronged patient engagement strategies.
• Promote diversity of people and needs in HTA practice and patient engagement • Lead inclusive patient engagement • Provide practical support to patients • Raise awareness about patient engagement in HTA	• Leverage the functions of digital technologies to identify, recruit and engage patients with different needs and abilities • Address the digital divide

a) Build organizational capacity for inclusive patient engagement processes

The findings suggested that HTA organizations should build infrastructure to support inclusive patient engagement processes. HTA organizations could evaluate their operations to identify improvement areas and implement the necessary changes to make their engagement processes more inclusive. For example, HTA organizations could reinforce initiatives to ensure that underrepresented groups can participate in HTA. Research participants highlighted that offering travel accommodations to patients and leading dynamic patient engagement were positive steps towards more inclusivity in patient engagement.

HTA recommendations are not necessarily focused on impacting societal issues, like stigma and mistrust in healthcare systems. Still, HTA organizations can implement initiatives to improve public trust in the HTA process. Research participants suggested that increasing cultural competencies among HTA analysts and raising awareness about patient engagement in public could boost confidence in HTA. In addition to increasing the diversity of patients, the interviewees advised that HTA organizations be staffed with employees with different abilities and backgrounds to reflect the diversity of patients impacted by HTA recommendations. Other studies have also reported that diversity in staff can help increase public trust in research organizations (Gill et al., 2018; Reynolds et al., 2021).

b) Use digital technologies in multi-pronged patient engagement strategies

Digital technologies present great opportunities for promoting the participation of patients with diverse backgrounds in HTA. The interviewees appreciated the various advantages digital technologies offered in reducing barriers to participation in engagement for certain population groups. The COVID-19 pandemic experience has boosted the development of online patient communities. As they continue to grow, HTA organizations should tap into those communities to help increase the diversity of patients contributing to HTA. Digital technologies can support diversity and inclusivity at all stages of the patient engagement process.

For example, HTA organizations can use social media to connect with online patient communities and influencers to recruit and engage digitally savvy patients, especially those living with disabilities. Interactive functions of digital technologies can provide more flexibility for accommodating people with different abilities, such as patients with autism. Interconnectedness features can also help manage the emotional burden generated by reactions to HTA recommendations. It can further support public education about the roles of patient representatives as more diverse patients could observe HTA advisory committee sessions. HTA organizations can use asynchronous and synchronous features to let patients share their experiences to establish emotional connections with decision-makers.

Despite the advantages of digital technologies mentioned above, in-person options should remain available to patients to not further exclude individuals already digitally disadvantaged in society. Some people may also prefer in-person meetings because they believe meeting face-to-face provides more opportunities for richer communication than online interactions. The research participants consistently raised the digital divide issue in the interviews. Besides access challenges, manipulating digital devices, navigating spaces, and experiencing harassment, the virtual environment might deter some people from using digital technologies (Graetz et al., 2016; Lai & Widmar, 2020; Raghuram et al., 2019). Finally, HTA organizations should create a safe space that allows equitable and productive patient engagement activities (Karl et al., 2022).

c) Engage patients to clarify the meaning of health equity in HTA

Finally, the interviewees said that researchers need to work with patients to identify which health equity factors are more important to consider for improving health outcomes. Similar to previous studies, the qualitative study highlighted the need to find alignment among patients and researchers around the meaning of health equity (Dawson et al., 2018; Eslava-Schmalbach et al., 2017; Roche et al., 2020).

6.6.4. Conclusion

The qualitative study helped capture patient perspectives on designing patient engagement to develop equity-focused HTA recommendations in this digital era. The respondents highlighted the importance of their input for HTA recommendations; however, they also suggested that the current health equity analysis did not always capture their concerns. HTA organizations need to work with patients to establish how health equity could be better applied to their lived experiences while considering its implications for implementing HTA recommendations.

Notwithstanding this knowledge gap, the interviewees described digital technologies as valuable tools to support patient engagement designed to inform health equity analysis in HTA. They explained how HTA organizations should use digital technologies to help increase the inclusion of patients with different abilities, backgrounds, and needs in HTA. The respondents said that HTA organizations can address the digital divide by providing in-person engagement options and IT support.

Chapter 7: Manuscript 3

Title: HTA analysts ‘perspectives on patient engagement to inform equity-focused health technology assessment (HTA) recommendations in the digital era: a grounded theory case study

7.1. Overview of Chapter 7

Chapter 7 is a complete manuscript describing the qualitative study with HTA analysts. The manuscript covers the dissertation's second objective, identifying implementation considerations for patient engagement in HTA. Chapter 7 contains the following sections: abstract, background, methods, results, and discussion.

Targeted journal: International Journal of Technology Assessment in Health Care (IJTAHC)

Authors’ contributions:

- Rosiane Simeon is the Ph.D. candidate and lead author of the manuscript. Rosiane led the planning, research design, data collection, analysis, and manuscript writing.
- Peter Tugwell and Vivian A. Welch co-supervised the Ph.D. candidate and contributed to the planning, research design and review of the manuscript.

- Ian D. Graham, Kumanan Wilson, Reiner Banken, Shehzad Ali, and Janet H. Roberts are the Thesis Advisory Committee (TAC) members and contributed to the research design and review of the manuscript.
- Anita Rizvi (AR): is a co-author and contributed to data analysis and interpretation of findings.

7.2. Abstract

Background: Health technology assessment (HTA) is a knowledge creation activity where HTA analysts assess evidence and engage different stakeholder groups in developing policy recommendations. An equity-focused HTA recommendation balances the benefits and harms of health technologies to help reduce the burden of adverse health outcomes on individuals who are disadvantaged in society. However, it is unclear how current patient engagement strategies inform health equity analysis in HTA.

Objective: To identify implementation considerations for patient engagement to inform health equity analysis in two Canadian HTA organizations: the Canadian Agency for Drugs and Technologies in Health (CADTH) and Health Quality Ontario (HQO).

Methods: We interviewed six English-speaking HTA analysts who worked in CADTH and HQO. We used a combination of grounded theory and thematic analysis to analyze the interviews and to guide the interpretation of findings.

Results: Qualitative analysis of interviews revealed a need to tailor current patient engagement strategies to enhance their impact on developing equity-focused HTA recommendations. The research participants said that CADTH and HQO should consider using non-digital and digital tools to ensure patients from diverse backgrounds can contribute to health equity analysis in HTA.

Conclusions: These findings can inform the design and evaluation of patient engagement for health equity analysis in HTA.

7.3. Introduction

Health technology assessment (HTA) is a knowledge-creation activity where HTA analysts assess evidence and engage different stakeholder groups in developing recommendations for policymakers (Wahlster et al., 2017). The HTA process comprises several stages including the identification of health technology for assessment, framing of the scope of HTA, review of the evidence, conduct of stakeholder engagement and reach of deliberative decision-making to develop HTA recommendations (Wahlster et al., 2017a). An equity-focused HTA recommendation balances the benefits and harms of health technologies in ways that reduce the burden of adverse health outcomes on individuals who are disadvantaged in society (Benkhalti et al., 2021; Braveman & Gruskin, 2003; V. Welch et al., 2008). Patient engagement in HTA involves a collaboration between HTA organizations and patients to capture diverse perspectives about health technologies (Facey Helle et al., 2017; Manafo et al., 2018). However, it is only one approach to including health equity factors in HTA (Cookson et al., 2017; Espinoza & Cabieses, 2014).

Processes of patient engagement in population health, including in HTA, are increasingly gaining attention (Biddle et al., 2021; Davis et al., 2019; de Wit et al., 2020; Domecq et al., 2014; Facey, 2019; Greenhalgh et al., 2019; Snow et al., 2018). A recent scoping review identified 14 models and frameworks of patient engagement in health-related research (Chudyk et al., 2022); there were 18 overlapping and 57 divergent elements across six conceptual categories (Chudyk et al., 2022). Considering this conceptual landscape, it becomes essential to identify which procedural factors best support the purpose of patient engagement (Chudyk et al., 2022; de Wit et al., 2020; Frank et al., 2020; Greenhalgh et al., 2019).

HTA organizations that operate in democratic systems may focus on patient engagement strategies that consider the diverse representation of patients to elucidate choices, utilization and fair distribution of health technologies (Bellemare et al., 2018; Cookson & Mirelman, 2017; Lopes et al., 2016; Manafo et al., 2018). In other political systems, HTA organizations may focus their patient engagement strategies on establishing consensus around the use and coverage of health technologies (Coulter, 2012; Espinoza & Cabieses, 2014; Shams Moattar et al., 2016). Patients continue to demand more accountability from health authorities and call for consideration of their voices in decisions regarding their health (Snow et al., 2018; Staley, 2015). Studies also showed a need to increase the diversity of patients who contribute to HTA(Black et al., 2018; de Wit et al., 2020; Snow et al., 2018).

Studies described diversity in patient engagement as including less traditional voices in health research (Dawson et al., 2018; Khuntia et al., 2022; Roche et al., 2020). These less traditional patients' voices may consist of people underrepresented in research because of barriers regarding to their living conditions, geographical locations, physical and mental health conditions, socio-economic status, and lower literacy levels (Gill et al., 2018; Reynolds et al., 2021; Roche et al., 2020). Underrepresented population groups in health research may also include people who avoid the health systems because of traumatic experiences related to stigma and systemic barriers to accessing healthcare(Davis et al., 2019; Krieger, 2014).

Researchers have resorted to several strategies to address issues surrounding diversity and representativeness in patient engagement (Rowland & Kumagai, 2018; Safaei, 2015). Some researchers used democratic rights as the guiding principle for engaging patients in studies (Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018). A patient representative is, therefore, selected through either nomination or election to contribute to patient engagement on behalf of other patients (Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018).

Another approach is to consider statistical representation, which implies that a patient can relay the average experience of all patients with the same condition (Rowland & Kumagai, 2018). An obvious issue with this approach is the assumption that the patient representative has the knowledge and skills to convey the experiences of the whole patient group (Crockett et al., 2019; Mazor et al., 2016; Rowland & Kumagai, 2018). Other researchers used symbolic representation, focusing on patients' unique experiences to bring individual perspectives in developing research narratives (Dukhanin et al., 2020; Rowland & Kumagai, 2018). From this viewpoint, a patient representative shares their story as their own authentic and unique experience, which may not necessarily reflect the lived experiences of other people with the same condition (De Santis et al., 2019; Fischer & Van de Bovenkamp, 2019; Rowland & Kumagai, 2018).

Digital technologies continually transform how patients interact with their environment (Doupi, 2016; Kent & Yellowlees, 2015; Mitchell & Kan, 2019). Healthcare systems are increasingly adopting digital technologies for mainstream activities to capture patient insights (Alami et al., 2020; Brewer et al., 2020; Mitchell & Kan, 2019; Wilson & Flood, 2021). It is critical to identify organization-specific factors to leverage the inclusion of patients from diverse backgrounds and needs to inform the development of equity-focused HTA recommendations.

7.4. Objective and research questions

The qualitative case study aims to identify implementation considerations for patient engagement to inform the development of equity-focused HTA recommendations. This case study addressed the following research questions:

- How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?
- How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?

7.5. Research design and methods

We described We described the research design, methods, and data analysis in Chapter 6.

7.6. Findings

7.6.1. Characteristics of participants

RS identified potential research participants by contacting the HTA agencies. RS approached a total of eight HTA analysts for this study. Two of them did not respond. RS interviewed six English-speaking HTA analysts who worked at CADTH and HQO. The research participants had six to twenty years of experience working in the field of HTA. We were not allowed to report on the positions and responsibilities of research participants to protect their privacy and identity. In similar studies (Malterud et al., 2016; Nelson, 2017), interviewing three participants in the same organization was sufficient to answer the research questions.

7.6.2. Synthesis and interpretation

In this section, we described the perspectives of six HTA analysts in CADTH and HQO. RS used a grounded theory and thematic analysis approach to analyze the interviews. Then, RS used a conceptual framework composed of items from the knowledge-to-action (KTA) cycle and the technology acceptance model (TAM) to organize the previously identified themes. We summarized the key findings in **Table 9**. First, we reported the interviewees' strategies for patient engagement to enhance health equity analysis in HTA. Then, we described the drivers of implementing patient engagement to inform health equity analysis in HTA. Finally, we discussed the interviewees' recommendations for implementing patient engagement to inform the development of equity-focused HTA recommendations. To manage the length of the manuscript, we reported illustrative quotes in **Appendix F**.

Table 9: Summary of key findings

Themes	Subthemes
Q1: How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?	
1. Current strategies for patient engagement to enhance health equity analysis in HTA	• Ensure diverse representation ○ Statistical and symbolic representation • Collaborate with grassroots organizations • Use multiple communication modalities • Lead dynamic engagement
2. Drivers of implementing patient engagement to inform health equity analysis in HTA	• Data availability • Culture of evidence in healthcare • Learning curve and training • Characterizing and reporting health equity factors • Implications of developing equity-focused HTA recommendations • Funding for patient engagement activities • Compensation policy • Mistrust in health systems and government • Public awareness about patient engagement
Q2: How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?	

3. Considerations for implementing digitally enabled patient engagement strategies for equity-focused HTA recommendations	• Use of digital technologies to:
	  ○ Increase reach
	  ○ Increase social interactions.
	• Consider the digital divide
	  ○ Provide in-person engagement options

Theme 1: Patient engagement strategies for health equity in HTA

In this section, we reported on research participants' strategies to implement patient engagement to enhance health equity analysis in their organizations. The interviewees recognized the importance of patient engagement to inform the development of equity-focused HTA recommendations. They expressed that patients' lived experiences helped bring personal and unique perspectives on implementing health technologies. They also stressed the importance of promoting diversity and inclusion in their work.

The research participants stated that they are already applying several strategies to help enhance the impact of patient engagement in health equity analysis in HTA. They described the following approaches when engaging patients in HTA: selecting patients with diverse backgrounds, partnerships with grassroot organizations, flexible engagement modalities, and dynamic engagement sessions.

Subtheme 1: Diverse and inclusive patient engagement

Statistical representation: Some interviewees explained that they used data in clinical trials to guide the selection of patients for informing HTA. They reported trying to recruit patients most affected by the conditions or health technologies they are assessing.

Symbolic representation: Another interviewee explained that diverse representation might be challenging in clinical trials and patient input. They said they must rely on patient organizations and advocates to help fill the knowledge gap. They explained that the commitment of patient representatives could also help bring health equity issues forward on behalf of those patients who may not have the opportunity to participate in patient engagement.

Subtheme 2: Partnerships with grassroots organizations

The HTA analysts felt that partnering with community organizations was crucial for recruiting patients who do not typically engage with the government because of trust issues. With varying degrees of success, these partnerships helped to connect with people at higher risk of experiencing adverse health outcomes.

Subtheme 3: Flexible engagement modalities

The research participants used multiple modalities to support patient engagement. They explained that having flexibility in communicating with patients helped them build trust. It also helped ease anxiety arising from patients' interaction with HTA analysts. An interviewee explained that meeting patients where they are can make the engagement process more inclusive and equitable while helping to understand better the context of implemented health technologies.

Digital modalities: The telephone is the default modality for interviewing patients; email is typically used to exchange documents.

Physical mailing: An interviewee reported using physical mailing addresses to communicate with patients because the latter may be comfortable with using emails or a printer.

In-person meeting: Another interviewee said they had met patients at their homes because they may have a disability that prevents them from manipulating a telephone. Other patients are maybe more comfortable meeting in their community.

Focus groups: An interviewee reported using focus groups to engage patients, especially those living in remote areas.

Subtheme 4: Dynamic engagement session

Clear communication: The interviewees have highlighted clear communication as a critical factor for successful patient engagement. They said it is essential to outline the feedback HTA organizations seek from patient groups. For example, resources that provide guidance on the content and support for a successful patient input submission should be available to patient groups.

Show respect and empathy: They also explained that showing respect through active listening during patient engagement sessions can increase patients’ confidence to share their experiences. They discuss that active listening can help show empathy and demonstrate that the ultimate role of HTA analysts is to help patients beyond all the bureaucracy involved in conducting patient engagement.

Theme 2: Drivers of patient engagement to inform health equity analysis in HTA

In **Table 10**, we summarized drivers for conducting patient engagement to inform the development of equity-focused HTA recommendations We discussed research participants' perceptions of challenges and solutions regarding these drivers in the paragraphs below.

Table 10: Drivers of patient engagement for health equity

Challenges	Proposed solutions
• Scoping patient engagement o Data availability o Culture of evidence in healthcare • Interest of the public o Mistrust in government o Lack of awareness • Organizational capacity for patient engagement o Timeline of HTA o Funding o Compensation policy • Health equity analysis in HTA o Characterizing and reporting health equity factors o Learning curve o Implications of developing equity-focused HTA recommendations	• Require disaggregated data in clinical trials • Promote patient engagement in HTA • Collaborate with patient advocates • Raise public awareness about patient engagement • Compensate patients • Collaborate with policymakers • Train HTA analysts

Subtheme 1: Data availability and culture of evidence in healthcare

Data availability: One interviewee explained that it is difficult for HTA analysts to decide on developing health equity questions because of a lack of data in clinical trials. They explained that original studies do not systematically report disaggregated data on population characteristics. Another interviewee suggested that regulatory bodies should require pharmaceutical companies to conduct subgroup analyses.

Culture of evidence in healthcare: An interviewee explained that some of their colleagues accord more value to quantitative data than qualitative data, like patient input. They suggested that the tension around the scientific value of qualitative versus quantitative data in healthcare affect interest in patient engagement in HTA. Another research participant said HTA analysts must collaborate to promote patient engagement in developing HTA recommendations.

Subtheme 2: Interest of the public in patient engagement

Mistrust in government and healthcare: The interviewees expressed that mistrust in government and healthcare systems is a significant barrier for some individuals to participate in patient engagement. They said some patients do not trust HTA analysts because they associate HTA organizations with the government. The interviewees suggested that HTA organizations need to develop collaboration with local organizations and community leaders to help connect with patients.

*Awareness about patient engagement in public***:** The interviewees explained that the pool for recruiting patients is often restricted to people with frequent contact with the healthcare systems. In addition, the research participants said that some people might not even be aware of the opportunity to participate in patient engagement. Other individuals may not take a genuine interest in patient engagement because they do not see any value in it. For these reasons, some interviewees recommended HTA organizations implement innovative campaigns to persuade the public about the role of patient engagement in shaping healthcare systems.

Subtheme 3: Organizational capacity for patient engagement to inform health equity

Funding for patient engagement activities: Some interviewees discussed the impact of the lack of funding for patient engagement activities in HTA. One interviewee explained that patient organizations might need funding to lead research to gather input that can elicit health equity concerns amongst their members. Another interviewee discussed that HTA organizations might need to allocate more budget to increase the number of HTA analysts doing patient engagement.

Compensation policy: An interviewee reported their HTA organization does not have the policy to compensate patients for their contribution to the HTA process. They suggested that paying patients for their time could help increase diversity and inclusion in patient engagement. For example, providing compensation could help improve the representation of patients in a lower income range.

Timeline of HTA: Some interviewees explained that the timeframe of HTA does not allow for conducting a comprehensive analysis of patient input. They explained that more time analyzing patient information could help identify more equity factors.

Training: Some interviewees also highlighted the need for HTA organizations to train HTA analysts in implementing patient engagement that can elicit health equity concerns.

Subtheme 4: Health equity analysis in HTA

Characterizing and reporting health equity factors: The interviewees expressed that the lack of standardization in reporting health equity factors in HTA is a challenge. Some interviewees said that they conducted a health equity analysis but did not necessarily report it. Another interviewee explained that HTA analysts might encounter challenges finding the accurate language to describe health equity issues. Some interviewees articulated how the mandates of patient organizations could affect identifying and reporting health equity factors. They explained that HTA organizations focusing on treatment access might identify more health equity factors than HTA organizations providing peer support.

Use of conceptual frameworks: Some research participants suggested HTA organizations systematically use conceptual frameworks to integrate patient input into HTA recommendations. Other interviewees highlighted the limitations of using social-determinant-based frameworks for eliciting health equity factors in patient input. They provided examples where these frameworks could not help to describe health inequities arising from the design of health technologies.

Learning curve: Given all the arguments surrounding health equity, some interviewees suggested that HTA analysts need time to learn about its application in HTA. An interviewee explained that HTA analysts do not fully understand the scope of health equity in HTA. Some interviewees cited circumstances where health equity analysis is unnecessary. Another interviewee argued that health equity is inherent to the HTA process and that HTA organizations must only report the rationale of their health equity decisions.

A research participant said that HTA analysts may have identified health equity concerns, but they may not know how to document choices to develop HTA recommendations. Another interviewee explained that HTA analysts might not know at which stage of the HTA process to apply health equity. Some interviewees also provided examples of conducting health equity analysis without patient engagement.

Implications of developing equity-focused HTA recommendations: An interviewee reported that HTA analysts may have identified equity factors, but those concerns may not make it to the HTA recommendations. Another interviewee discussed that some of the equity-focused recommendations fall outside the realm of the healthcare systems.

It is difficult for HTA analysts to determine which jurisdictions are responsible for implementing those non-healthcare-specific recommendations. Some interviewees suggested that

HTA organizations must work with policymakers to clarify the purpose of developing equity-focused recommendations.

Theme 3: Considerations for patient engagement to inform health equity in this digital era

We used the technology acceptance model (TAM) to describe considerations for implementing digitally-enabled patient engagement to inform health equity in HTA. As a reminder, the TAM suggests that the intention to use a technology depends on users' perceptions of the usefulness and ease of utilizing the technology. Then we used the KTA cycle to organize the interviewees' recommendations into practical and ethical considerations for implementing the suggested strategies.

In general, the research participants believed that the strategic use of digital could help implement patient engagement that can lead to equity-focused HTA recommendations. The interviewees also shared some caveats regarding the digital divide. In addition to considering the features of digital tools, HTA organizations must factor in patients 'social environment and personal preferences when developing digitally-enabled patient engagement strategies. We summarized the considerations for using digital technologies for diverse and inclusive patient engagement in **Table 11**.

Table 11: Considerations for digitally-enabled patient engagement.

Practical considerations	Ethical considerations
Perceived usefulness • Increase reach. • Increase social interaction. • Conduct social media listening.	Perceived ease of use • Have access to digital technologies. • Prefer virtual or in-person meetings. • Can manipulate digital tools. • Feel safe in a virtual environment.

Subtheme 1: Practical considerations

The research participants said that the strategic use of digital technologies could help address some practical issues to make patient engagement more diverse and inclusive.

Increase reach and social interaction: The interviewees described digital technologies as a practical means for HTA organizations to expand their reach. They believed that the COVID-19 pandemic has made digital technologies more relevant and widely acceptable for conducting patient meetings. They described the practicality of embedded interactive and transcription features in some digital solutions popularized during the COVID-19 pandemic.

Social media outreach and monitoring: Some interviewees voiced how HTA organizations could leverage social media to advertise engagement opportunities. Another interviewee suggested that HTA organizations could consider using social media monitoring to inform the scope of patient engagement. They said social media could offer rich perspectives on patient experiences.

Subtheme 2: Ethical considerations

The research participants have stressed the importance of considering ethical issues related to privacy and the digital divide when deploying patient engagement strategies. Adopting digital technologies without considering their attributes could lead to further inequity in patient engagement.

Privacy issues: The HTA analysts we interviewed emphasized the importance of implementing ethical procedures to reassure patients in virtual engagement. One interviewee suggested that HTA organizations should be transparent with patients engaged in their process to help manage expectations about using information and privacy.

Digital divide: Research participants highlighted the need to consider the digital range for the systematic use of digital technologies for patient engagement, including social conditions, education, and digital literacy. In addition to access, debilitating conditions could prevent some individuals from using essential digital technologies such as a telephone.

Subthemes 3: Mixed communication modalities

The interviewees warned that issues of access and affordance around digital technologies could amplify inequity in patient engagement. Therefore, they recommended that non-digital tools be used alongside digital technologies to avoid further exclusion of those affected by the digital divide

Digital technologies: As explained above, research participants recognized the potential of digital technologies to leverage current strategies for diverse and inclusive patient engagement.

Electronic and physical mailing: An interviewee explained how emails and web portals conveniently exchange documents between HTA organizations and patients. Still, some patients have opted for receiving documents by mail because they may not have access to a printer or do not know how to use an electronic signature.

In-person meetings: Another interviewee further articulated how in-person meetings allow for more genuine conversations, better understanding, and richer data than virtual engagement.

7.7. Discussion

7.7.1. Summary of Findings

We used a grounded theory approach and thematic analysis to analyze the perspectives of six HTA analysts from CADTH and HQO on strategies to improve patient engagement to inform the health equity analysis in HTA. The HTA analysts shared about current practices that allowed them to successfully reach out, recruit and engage underrepresented patient groups in their respective institutions. The research participants have recognized the role of digital technologies in strengthening organizational readiness for patient engagement to incorporate equity factors in HTA recommendations. We summarized the study's findings in the following paragraphs.

a) Patient engagement strategies to inform health equity in HTA

As in similar studies (Black et al., 2018; de Wit et al., 2020; Snow et al., 2018), the interviewees acknowledged the importance of patient engagement as evidence to help fill gaps identified in clinical and economic data to inform the development of HTA recommendations. They described initiatives in their respective organizations to foster active participation and broaden the representation of patients with diverse backgrounds in HTA. The research participants reported they used several strategies in patient engagement, including targeted outreach, development of partnerships, multiple communication modalities, and valuing patient input. The HTA analysts, however, described mistrust in government, lack of awareness about the value of patient engagement and lack of compensation to patients as critical challenges to implementing the strategies mentioned above.

They recommended that HTA organizations raise public awareness of the value of patient engagement and put compensation policies in place to support it. They also reiterated the need to collaborate with community leaders to help increase the recruitment of underrepresented patients in HTA. They further highlighted the role of patient advocacy, both individually led or through patient organizations, as an enabler for identifying health equity factors to inform HTA recommendations. Some research participants, however, believed that the mandate of patient organizations plays a role in identifying and reporting health equity factors in patient input.

The above accounts are aligned with earlier studies which showed that patient engagement that considered the diversity of people affected by health interventions led to capturing health equity concerns to inform health policies (Jull et al., 2018; Snow et al., 2018). Considering the above findings, working closely with community-based organizations could help increase trust in HTA organizations while encouraging the participation of underrepresented groups. Recognizing that trust building takes time, such initiatives could help enhance the representation of patients and may eventually lead to health equity in HTA. Further research could also help elucidate assumptions and the extent to which patient mandates may have impacted the integration of health equity factors in HTA recommendations.

b) Considerations for implementing digitally-enabled patient engagement

The research participants believed digital technologies could support efforts to create conducive environments for inclusive patient engagement. They have identified practical and ethical considerations for leveraging digital technologies. They expressed that digital technologies could help reduce logistic barriers, increase social interaction during engagement sessions and identify patient perspectives through social media monitoring. They have also discussed how relying solely on digital technologies for patient engagement can further contribute to the digital divide.

Digital technologies such as interactive applications and mobile devices that require reliable access to the internet and manipulations might not be as widely used as phones. Sometimes, in-person meetings might be the only option to engage patients. Some patients may be unable to use a phone; others may not have access to email and printers. Because of the above ethical reasons, interviewees seemed to favour a balanced approach to implementing digitally-enabled patient engagement for HTA. The research participants explained that HTA organizations should consider using in-person and non-digital means to sustain patient engagement. By doing so, HTA organizations would mitigate ethical issues arising from the accessibility and affordance of digital technologies.

Based on the above findings, the research participants agreed that digital technologies provide more opportunities to involve patients from diverse backgrounds in HTA. However, systematically considering the social aspects surrounding digital technologies can help tease out equity issues that may arise during their deployment for patient engagement. CADTH and HQO should consider using non-digital and digital tools to ensure that all patients can contribute to developing equity-focused HTA recommendations.

7.7.2. Strengths and limitations

We used a combination of grounded theory and thematic analysis to develop study findings. We also focused on addressing previously identified issues in the organizations of interest. The study results are context-bound regarding knowledge translation and maybe more pertinent for CADTH and HQO than other HTA organizations in Canada and elsewhere. Even in those organizations, there may be a need for further research around the impact of patient organization mandates on reporting health equity concerns in patient input. The findings can also inform planning for implementing digitally-enabled patient engagement to inform health equity in similar organizations.

7.7.3. Implications for practice, policy, and research

The findings of this qualitative study can serve as a rationale for evaluating current patient engagement strategies in CADTH and HQO. Such assessments may lead to developing novel solutions that integrate new digital technologies in an overarching strategy to enhance organizational capabilities to collect and analyze input from patients with diverse backgrounds. In addition, the findings can inform training strategies for HTA analysts and revision of the HTA timeline. The results can guide the collaboration between HTA organizations and policymakers to help clarify the purpose of HTA recommendations in advancing health equity in their jurisdictions. Further research can explore the association between the mandates of patient organizations and the reports of health equity factors in patient input.

7.7.4. Conclusion

In this explanatory case study, we used a combination of grounded theory and thematic analysis to categorize findings based on the knowledge-to-action (KTA) framework and the technology acceptance model (TAM). The study revealed that HTA organizations must consider critical practical and ethical considerations to support patients with diverse backgrounds in patient engagement. HTA organizations should weigh the usefulness of digital technologies for accessing a diverse patient audience and sustaining inclusive engagement. HTA analysts should also ensure that the characteristics of digital technologies are adapted to patients' social environments while protecting the privacy and safety of those participating in virtual engagement.

Digital technologies could support engagement to inform health equity while helping streamline the patient input collection and analysis process. A thorough evaluation of patient engagement may help to expose potential flaws in current patient engagement processes for incorporating health equity factors in HTA recommendations. While insights from this research are drawn from two Canadian HTA organizations evolving in two different contexts, they may also echo the needs of other HTA institutions focused on advancing health equity.

Chapter 8: Integrated discussion

8.1. Overview of Chapter 8

Chapter 8 includes an integrated discussion of the dissertation findings. First, I provide a summary of each study. Second, I present the main points covered in the integrated discussion. Third, I discuss the strengths and limitations of the dissertation overall. I discussed disciplinary implications for practice, education, policy, and research. I conclude the chapter with a summary of the results and their implications for HTA organizations, patients, researchers, and decision-makers.

8.2. Summary of main findings from the dissertation

I provide a summary of each study in **Table 12**, **Table 13**, and **Table 14**. The thesis examines the association between patient engagement processes and equity-focused HTA recommendations and identifies implementation considerations for patient engagement in HTA. I address the objectives by answering the following research questions:

- What are the characteristics of patient engagement processes in HTA?
- What are the characteristics of equity-focused HTA recommendations?
- What patient engagement processes are associated with equity-focused HTA recommendations?
- How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?
- How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?

Table 12: Chapter 5-Summary of findings.

Objective: Examine the association between patient engagement and equity-focused HTA recommendations		
Q1: What are the characteristics of patient engagement processes in HTA?	Q2: What are the characteristics of equity-focused HTA recommendations?	Q3: What patient engagement processes are associated with equity-focused HTA recommendations?
• Patients contributed to HTA as key informants and members of advisory committees. • Patient organizations generally use more diverse methods to engage patients than HTA organizations. • In HQO HTAs, HTA analysts directly interview patients to collect their input, and the advisory committee uses consensus as the default decision-making model. • In pCODR HTAs, Patient groups submit patient input on behalf of their members to CADTH, and the advisory committee uses consensus as the default decision-making model. • In CDR HTAs, Patient groups submit patient input on behalf of their members to CADTH, and the advisory committee uses vote as the default decision-making model.	• Top five PROGRESS-Plus items reported in HTA recommendations: 1. Affordability 2. Logistics of treatment 3. Place of residence 4. Socioeconomic factors 5. Severity of conditions • HQO reviews had the highest proportion of HTA reports with equity-focused recommendations. • There were any of the following items from the PROGRESS framework-occupation, race/ethnicity/culture, and religion- in any sections of the included HTA reports • The following PROGRESS-Plus items were inpatient input only: stigma, social capital, and gender.	• Hypothesis: Direct patient engagement or consensus in decision-making is more likely to lead to equity-focused HTA recommendations than indirect engagement or vote. • Direct engagement (OR: 3.85; 95% CI: [2.40 – 6.20]) and consensus in decision-making (OR: 2.27; 95% CI: [1.35 – 3.84]) were more likely to be associated with reporting health equity factors in HTA recommendations than indirect engagement (OR: 0.26; 95% CI: [0.16 – 0.41]) and voting (OR: 0.44; 95% CI: [0.26 – 0.73])

Table 13: Chapter 6- Summary of findings.

Objective: Identify implementation considerations for patient engagement to inform the development of equity-focused HTA recommendations	
Q1: How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?	Q2: How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?
The qualitative analysis of interviews with patients revealed that HTA organizations should: • Engage patients to clarify the meaning of health equity. • Promote diversity of people and needs in HTA practice and patient engagement. • Lead dynamic and meaningful engagement. • Build organizational capacity to implement inclusive patient engagement.	The qualitative analysis of interviews with patients revealed that HTA organizations could: • Use digital technologies in multi-pronged strategies to enhance the contribution of patients with diverse backgrounds and needs in HTA. • Consider in-person engagement to compensate for the digital divide.

Table 14: Chapter 7- Summary of findings.

Objective: Identify implementation considerations for patient engagement to inform the development of equity-focused HTA recommendation	
Q1: How could HTA organizations ensure that patient engagement effectively influences the development of equity-focused HTA recommendations?	Q2: How could HTA organizations use digital technologies to engage patients in ways that acknowledge the diversity of people and needs impacted by health technologies?
The qualitative analysis of interviews with HTA analysts showed that HTA organizations need to: • Promote a culture of evidence that values patient input. • Increase collaboration with community organizations. • Raise awareness about patient engagement in the public • Build strategies to support patient engagement processes that can inform equity-focused HTA recommendations.	The qualitative analysis of interviews with HTA analysts showed that: • HTA organizations could use digital technologies to help: o Increase reach o Promote social interactions o Implement social media outreach and social media listening • HTA organizations should consider the digital divide, including issues related to access to digital technologies, communication modalities preferences and safety in virtual environments.

8.3. Main points of integrated discussion

The findings suggested that patient engagement may be critical for identifying health equity factors in HTA. The dissertation also revealed that factors such as the context of HTA, methods of collecting patient input and model of decision-making in the HTA advisory committee could mediate this relationship. Patients and HTA analysts interviewed for the dissertation have reiterated the importance of patient engagement for identifying health equity concerns related to implemented health technologies and the burden of illness. They have also identified organizational challenges that impede the contribution of patients with diverse backgrounds and abilities in developing equity-focused HTA recommendations.

The HTA analysts interviewed have also highlighted conceptual and practical challenges encountered in implementing patient engagement to inform health equity analysis in HTA. Previous studies have corroborated research participants' concerns about conceptual frameworks for patient engagement. They revealed little guidance on increasing the inclusion of marginalized population groups in health research (Greenhalgh et al., 2019; Roche et al., 2020). The interviewees have presented recommendations to HTA organizations for addressing the identified challenges. In the following paragraphs, I discussed the various considerations that emanated from the dissertation.

8.3.1. Frame patient engagement as an intervention to fit its purpose

Based on the dissertation's findings, it is essential to frame patient engagement as an organization-level intervention to enhance health equity analysis in HTA. When analyzing the relationships between patient engagement and HTA recommendations, we saw that the context of the HTA and procedural elements such as decision-making models could make a difference in the number of health equity factors in HTA recommendations. Consensus as a decision-making model seemed to play a role in increasing health equity factors in HTA recommendations; however, HTA organizations collected patient input. On the other hand, research participants in the qualitative study have stressed the need to improve current engagement processes for bringing the unique perspectives of patients in informing the development of equity-focused HTA recommendations.

The findings of this dissertation can serve as an evidence base for CADTH HQO and patient organizations to frame patient engagement as an intervention to support health equity analysis in HTA. Elucidating the context of HTA and procedural elements of patient engagement and interactions amongst these elements can help develop patient engagement strategies that fit their predefined purposes.

Challenges: Previous research on patient and public involvement found that existing frameworks failed to present a clear pathway for implementing patient engagement that can lead to the integration of health equity (Chudyk et al., 2022; Domecq et al., 2014; Greenhalgh et al., 2019). Therefore, HTA and patient organizations must work with relevant stakeholders to devise the most adapted processes to the context and purpose of their patient engagement. Such an approach may lead to patient engagement strategies pertinent to patients and HTA organizations. It can ultimately increase the diversity of people and perspectives to further advance health equity through HTA (Greenhalgh et al., 2019; Roche et al., 2020).

8.3.2. Build organizational capacity for patient engagement to inform health equity

The research findings have also exposed a need to build organizational capacity to enhance patient engagement's impact on health equity analysis in HTA. Research participants identified several areas of improvement to sustain patient engagement for health equity in HTA organizations. They cited the culture of evidence in healthcare, lack of funding for patient engagement activities, lack of cultural competencies amongst HTA researchers and lack of public awareness around the importance of patient engagement as critical challenges to implementing diverse and inclusive patient engagement.

Challenges: From an organizational readiness point of view, HTA organizations that directly conduct patient engagement can use the above rationale to initiate internal discussions about procedural issues for tailoring patient engagement to inform health equity analysis. However, it might be more difficult for HTA organizations that conduct engagement through patient organizations to take that route. These HTA organizations may not have much influence on patient organizations.

8.3.3. Use of digital technologies for patient engagement in HTA

The research participants, patients and HTA analysts, have expressed appreciation for the new digital features to implement patient engagement. Digital technologies will remain a key instrument in the patient engagement toolbox for well-established reasons such as reducing location constraints, costs associated with in-person meetings and management of social interactions (Jenssen et al., 2016; Liddy et al., 2017). Research participants have focused their attention on ways to optimize the use of digital technologies to support inclusive patient engagement.

For example, patient-participants mentioned that social media could help identify emerging topics around health technologies. They recognized that digital technologies could help solve critical logistics issues surrounding data collection and communication with patients with disabilities and those with different abilities that may need immediate adjustment during social interaction.

Challenges: The interviewees have advised against relying solely on digital technologies to promote diversity and inclusion in patient engagement. Patients and HTA analysts who participated in the study have stressed the need to factor in the digital divide in deploying patient engagement for health equity in HTA

8.3.4. Build organizational capacity for health equity analysis in HTA

The quantitative study showed that conceptual frameworks such as the PROGRESS-Plus and the equity consideration in HTA (ECHTA) checklist could facilitate identifying and reporting health equity factors in HTA. Some research participants have also used these conceptual frameworks in their work. However, HTA analysts and patients interviewed offered some caveats on using social-determinant-based frameworks for health equity in HTA. Some interviewees articulated that these frameworks did not perform as they expected for identifying inequities in the design of health technologies. In addition, some HTA analysts interviewed have raised conceptual issues related to the application of health equity in HTA. For example, some HTA analysts felt that health equity is new to HTA. Other HTA analysts argued that HTA is a response to health inequities; in this sense, all HTA recommendations address health equity. These conceptualization issues may have also impacted the importance of health equity analysis in HTA organizations.

Challenges: There is no consensus on the relationship between health equity and HTA. In that case, it might be challenging to establish a need for conducting a health equity analysis in the first place. Nevertheless, the research participants, including patients, recognized that addressing health equity is essential and recommended more consistency in reporting health equity considerations in HTA recommendations.

8.4. Strengths and limitations of the dissertation

This dissertation has several strengths. First, I consulted HTA analysts and patients who had experiences in HTA in Canada to determine the dissertation's scope and inform the qualitative study protocol. Second, the research questions focused on identifying practice gaps in two Canadian HTA organizations operating at different government levels. This narrow focus of the dissertation is both a strength and a limitation. The focus on two organizations can facilitate using the findings in those organizations. Still, some results might apply in settings with similar mandates as CADTH or HQO.

I did not extract data in duplicate for any of the three studies. However, I draw upon validated conceptual frameworks to develop a data extraction form and codebook to guide data collection and analysis. I met with data extractors weekly to review the data extraction sheet for accuracy. I also used a validated conceptual framework to analyze and interpret qualitative data. I sent summaries of preliminary findings to research participants to help validate the analysis of interview data. Finally, I sought feedback from my thesis advisory committee (TAC) members, composed of academics with expertise in health equity, knowledge translation, HTA, and digital technologies for health promotion.

8.5. Disciplinary implications

The dissertation findings have implications for practice, policy, education, and research.

8.5.1. Implications for practice

These findings can inform strategies around patient engagement and health equity in HTA with implications for HTA organizations, patient organizations, and patient advocates. In the short term, HTA organizations can use these findings to initiate evaluation around patient engagement practices and identify procedural parameters in current processes to enhance the impact of patient engagement in health equity analysis.

From a long-term standpoint, the findings can inform the design of patient engagement as an intervention embedded in corporate planning to advance health equity. For example, HTA and patient organizations can adjust their budget to include in-person and digitally-enabled options for patient engagement activities. HTA organizations could also use these findings to inform staff training on designing and facilitating inclusive patient engagement. Regarding health equity analysis, the results can inform training and knowledge exchange activities aimed at increasing the skills of HTA analysts.

Also, HTA organizations can work with patient advocates and organizations to roll out awareness campaigns to educate the public on the importance of patient engagement in HTA. HTA organizations, patient organizations and patient advocates can also use the findings of awareness about opportunities for the public to contribute to healthcare decisions through HTA. Given the challenge around the selection of patients, the findings can inform discussions around expectations of patient representation in HTA. Patient advocates can also use the results to inform their approaches to involvement in HTA processes.

8.5.3. Implications for policy

The dissertation findings also provide a basis for HTA organizations to develop a collaborative forum that brings together decision-makers and HTA analysts to help find alignment around the purposes of HTA recommendations with the types of information HTA organizations put in HTA recommendations. Alignment between the purpose and content of HTA recommendations can help inform strategies for developing HTA recommendations to advance health equity.

8.5.4. Implications for education

The dissertation findings have implications for education as it identifies training needs related to analytical skills for health equity in HTA. Academic programs in population health can aim to increase training in health equity analysis in HTA through continuing education for HTA analysts.

8.5.5. Implications for research

The dissertation findings showed that patients and HTA analysts encounter challenges in deciding on health equity factors to include in HTA recommendations. The field of HTA could benefit from further research to elicit a threshold of equity factors to consider in HTA recommendations. Research can cover questions outlined below about HTA practices for advancing health equity.

- What types of HTA models are more adapted to developing equity-focused HTA recommendations?
- What is the association between mandates of patient organizations (e.g., access to drugs versus peer support) and reporting of health equity factors in HTA recommendations?
- How do we conceptualize diversity and inclusion in patient engagement to support health equity in HTA?

8.6. Conclusions from the dissertation overall

In this article-based dissertation, I used an explanatory mixed methods approach to determine the relationships between patient engagement and the development of equity-focused HTA recommendations in two Canadian organizations. I interviewed HTA analysts and patients to identify considerations for tailoring patient engagement to inform health equity analysis in HTA. The findings suggested that patient engagement plays a role in integrating health equity factors in HTA recommendations. However, this association could be contingent on the decision-making models and types of patient engagement.

HTA analysts and patients interviewed for the research recommended that HTA organizations use digital and non-digital modalities to account for the digital divide, which may exclude some population groups from participating in patient engagement. I discussed the strengths, limitations, and implications of the findings. From a knowledge translation standpoint, knowledge users should consider the context of HTA organizations and health systems in applying the results to improve their patient engagement strategies for equity-focused HTA. In addition, policymakers should consider whether they have equity goals and resources to translate the study’s recommendations into policies. The dissertation results have paved the way for designing patient engagement strategies embodying organizational visions for advancing health equity

References

Adkins, E. M., Nicholson, L., Floyd, D., Ratcliffe, M., & Chevrou-Severac, H. (2017). Oncology drugs for orphan indications: How are HTA processes evolving for this specific drug category?. *ClinicoEconomics and Outcomes Research : CEOR, 9*, 327–342. https://doi.org/10.2147/CEOR.S134230

Allen, N., Liberti, L., Walker, S. R., & Salek, S. (2017). A comparison of reimbursement recommendations by European HTA agencies: Is there opportunity for further alignment? *Frontiers in Pharmacology, 8*(JUN). https://doi.org/10.3389/fphar.2017.00384

Allen, N., Walker, S. R., Liberti, L., & Salek, S. (2017). Health Technology Assessment (HTA) Case Studies: Factors Influencing Divergent HTA Reimbursement Recommendations in Australia, Canada, England, and Scotland. *Value in Health : The Journal of the International Society for Pharmacoeconomics and Outcomes Research, 20*(3), 320–328. https://doi.org/10.1016/j.jval.2016.10.014

AlQudah, A. A., Al-Emran, M., & Shaalan, K. (2021). Technology Acceptance in Healthcare: A Systematic Review. *Applied Sciences, 11*(22). https://doi.org/10.3390/app112210537

Aluwihare-Samaranayake, D. (2012). Ethics in Qualitative Research: A View of the Participants' and Researchers' World from a Critical Standpoint. *International Journal of Qualitative Methods, 11*(2), 64–81. https://doi.org/10.1177/160940691201100208

Angelis, A., Lange, A., & Kanavos, P. (2018). Using health technology assessment to assess the value of new medicines: Results of a systematic review and expert consultation across eight European countries. *European Journal of Health Economics*, *19*(1), 123–152. https://doi.org/10.1007/s10198-017-0871-0

Armstrong, M. J., Rueda, J.-D., Gronseth, G. S., & Mullins, C. D. (2017). Framework for enhancing clinical practice guidelines through continuous patient engagement. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *20*(1), Article 1. https://doi.org/10.1111/hex.12467

Ayouni, I., Maatoug, J., Dhouib, W., Zammit, N., Fredj, S. B., Ghammam, R., & Ghannem, H. (2021). Effective public health measures to mitigate the spread of COVID-19: A systematic review. *BMC Public Health, 21*(1), 1015. https://doi.org/10.1186/s12889-021-11111-1

Aziz, H., & Madani, A. (2015). *Evolution of the Web and its Uses in Healthcare*. http://qspace.qu.edu.qa/handle/10576/4067

Batalden, M., Batalden, P., Margolis, P., Seid, M., Armstrong, G., Opipari-Arrigan, L., & Hartung, H. (2016). Coproduction of healthcare service. *BMJ Quality & Safety*, *25*(7), 509. https://doi.org/10.1136/bmjqs-2015-004315

Beaunoyer, E., Dupéré, S., & Guitton, M. J. (2020). COVID-19 and digital inequalities: Reciprocal impacts and mitigation strategies. *Computers in Human Behavior, 111*, 106424. https://doi.org/10.1016/j.chb.2020.106424

Bellemare, C. A., Dagenais, P., K-Bedard, S., Beland, J.-P., Bernier, L., Daniel, C.-E., Gagnon, H., Legault, G.-A., Parent, M., & Patenaude, J. (2018). Ethics in health technology assessment: A systematic review. *International Journal of Technology Assessment in Health Care*, *34*(5), 447–457. https://doi.org/10.1017/S0266462318000508

Benkhalti, M., Espinoza, M., Cookson, R., Welch, V., Tugwell, P., & Dagenais, P. (2021). Development of a checklist to guide equity considerations in health technology assessment. *International Journal of Technology Assessment in Health Care*, *37*(gti, 8508113), e17. https://doi.org/10.1017/S0266462320002275

Biddle, M. S. Y., Gibson, A., & Evans, D. (2021). Attitudes and approaches to patient and public involvement across Europe: A systematic review. *Health & Social Care in the Community*, *29*(1), 18–27. https://doi.org/10.1111/hsc.13111

Bidonde, J., Vanstone, M., Schwartz, L., & Abelson, J. (2021). An institutional ethnographic analysis of public and patient engagement activities at a national health technology assessment agency. *International Journal of Technology Assessment in Health Care*, *37*(gti, 8508113), Article gti, 8508113. https://doi.org/10.1017/S0266462321000088

Black, A., Strain, K., Wallsworth, C., Charlton, S.-G., Chang, W., McNamee, K., & Hamilton, C. (2018). What constitutes meaningful engagement for patients and families as partners on research teams? *Journal of Health Services Research & Policy*, *23*(3), 158–167. https://doi.org/10.1177/1355819618762960

Bombard, Y., Baker, G. R., Orlando, E., Fancott, C., Bhatia, P., Casalino, S., Onate, K., Denis, J.-L., & Pomey, M.-P. (2018a). Engaging patients to improve quality of care: A systematic review. *Implementation Science*, *13*(1), 98. https://doi.org/10.1186/s13012-018-0784-z

Bombard, Y., Baker, G. R., Orlando, E., Fancott, C., Bhatia, P., Casalino, S., Onate, K., Denis, J.-L., & Pomey, M.-P. (2018b). Engaging patients to improve quality of care: A systematic review. *Implementation Science*, *13*(1), 98. https://doi.org/10.1186/s13012-018-0784-z

Bouchard, L., Albertini, M., Batista, R., & de Montigny, J. (2015). Research on health inequalities: A bibliometric analysis (1966-2014). *Social Science and Medicine*, *141*, 100–108. https://doi.org/10.1016/j.socscimed.2015.07.022

Boudes, M., Robinson, P., Bertelsen, N., Brooke, N., Hoos, A., Boutin, M., Geissler, J., & Sargeant, I. (2018). What do stakeholders expect from patient engagement: Are these expectations being met?. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *21*(6), 1035–1045. https://doi.org/10.1111/hex.12797

Bradley, E. H., Curry, L. A., & Devers, K. J. (2007). Qualitative data analysis for health services research: Developing taxonomy, themes, and theory. *Health Services Research*, *42*(4), 1758–1772. PubMed. https://doi.org/10.1111/j.1475-6773.2006.00684.x

Braveman, P., & Gruskin, S. (2003). Defining equity in health. *Journal of Epidemiology and Community Health*, *57*(4), 254 LP – 258. https://doi.org/10.1136/jech.57.4.254

Brewer, L. C., Fortuna, K. L., Jones, C., Walker, R., Hayes, S. N., Patten, C. A., & Cooper, L. A. (2020). Back to the Future: Achieving Health Equity Through Health Informatics and Digital Health. *JMIR Mhealth Uhealth*, *8*(1), e14512. https://doi.org/10.2196/14512

Brown, D. G., & Wobst, H. J. (2021). A Decade of FDA-Approved Drugs (2010–2019): Trends and Future Directions. *Journal of Medicinal Chemistry*, *64*(5), 2312–2338. https://doi.org/10.1021/acs.jmedchem.0c01516

Brüssow, H. (2013). What is health? *Microbial Biotechnology*, *6*(4), 341–348. https://doi.org/10.1111/1751-7915.12063

Busse, R., Panteli, D., Kreis, J., & Busse, R. (2015). Considering equity in health technology assessment: An exploratory analysis of agency practices. *International Journal of*

Technology Assessment in Health Care, *31*(5), 314–323. https://doi.org/10.1017/S0266462315000549

Callahan, D. (1973). The WHO definition of "health. *Studies - Hastings Center*, *1*, 77–88. https://doi.org/10.2307/3527467

Carey, T. S., Sanders, G. D., Viswanathan, M., Trikalinos, T. A., Kato, E., & Chang, S. (2012). *Taxonomy for Study Designs*. https://www.ncbi.nlm.nih.gov/books/NBK95280/

Carpiano, R. M., & Daley, D. M. (2006). A guide and glossary on postpositivist theory building for population health. *Journal of Epidemiology and Community Health*, *60*(7), 564–570. https://doi.org/10.1136/jech.2004.031534

Catarinella, F. S., & Bos, W. H. (2016). Digital health assessment in rheumatology: Current and future possibilities. *Clinical and Experimental Rheumatology*, *34*(5 Suppl 101), S2–S4. http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med12&NEWS=N&AN=27762198

Cavaller-Bellaubi, M., Faulkner, S. D., Teixeira, B., Boudes, M., Molero, E., Brooke, N., McKeaveney, L., Southerton, J., Vicente, M. J., Bertelsen, N., Garcia-Burgos, J., Pirard, V., Reid, K., & Ferrer, E. (2021). Sustaining Meaningful Patient Engagement Across the Lifecycle of Medicines: A Roadmap for Action. *Therapeutic Innovation & Regulatory Science*, *55*(5), 936–953. https://doi.org/10.1007/s43441-021-00282-z

Charlier, P., Coppens, Y., Malaurie, J., Brun, L., Kepanga, M., Hoang-Opermann, V., Correa Calfin, J. A., Nuku, G., Ushiga, M., Schor, X. E., Deo, S., Hassin, J., & Hervé, C. (2017). A new definition of health? An open letter of autochthonous peoples and medical anthropologists to the WHO. *European Journal of Internal Medicine*, *37*, 33–37. https://doi.org/10.1016/j.ejim.2016.06.027

Chudyk, A. M., Horrill, T., Waldman, C., Demczuk, L., Shimmin, C., Stoddard, R., Hickes, S., & Schultz, A. S. (2022). Scoping review of models and frameworks of patient engagement in health services research. *BMJ Open*, *12*(8), e063507. https://doi.org/10.1136/bmjopen-2022-063507

Chun Tie, Y., Birks, M., & Francis, K. (2019). Grounded theory research: A design framework for novice researchers. *SAGE Open Medicine*, *7*, 2050312118822927–2050312118822927. https://doi.org/10.1177/2050312118822927

Clark, B., & Preto, N. (2018). Exploring the concept of vulnerability in health care. *CMAJ*. https://doi.org/10.1503/cmaj.180242

Concannon, T. W., Grant, S., Welch, V., Petkovic, J., Selby, J., Crowe, S., Synnot, A., Greer-Smith, R., Mayo-Wilson, E., Tambor, E., Tugwell, P., & for the Multi Stakeholder Engagement (MuSE) Consortium. (2019). Practical Guidance for Involving Stakeholders in Health Research. *Journal of General Internal Medicine*, *34*(3), 458–463. https://doi.org/10.1007/s11606-018-4738-6

Concannon, T. W., Meissner, P., Grunbaum, J. A., McElwee, N., Guise, J. M., Santa, J., Conway, P. H., Daudelin, D., Morrato, E. H., & Leslie, L. K. (2012). A new taxonomy for stakeholder engagement in patient-centered outcomes research. *Journal of General Internal Medicine*. https://doi.org/10.1007/s11606-012-2037-1

Conference, H., & May, T. (2016). *Multi-stakeholder Approaches to Improve Evidence-Based Decisions in Rare Diseases: Engagement of Patients and Patient Organizations*. *May*, 1–15.

Cookson, R., & Mirelman, A. J. (2017). Equity in HTA: What doesn't get measured, gets marginalised. *Israel Journal of Health Policy Research*, *6*(1), 4–7. https://doi.org/10.1186/s13584-017-0162-3

Cookson, R., Mirelman, A. J., Griffin, S., Asaria, M., Dawkins, B., Norheim, O. F., Verguet, S., & J Culyer, A. (2017). Using Cost-Effectiveness Analysis to Address Health Equity Concerns. *Value in Health : The Journal of the International Society for Pharmacoeconomics and Outcomes Research*, *20*(2), 206–212. https://doi.org/10.1016/j.jval.2016.11.027

Cordoş, A.-A., Bolboacă, S., & Drugan, C. (2017). Social Media Usage for Patients and Healthcare Consumers: A Literature Review. *Publications*. https://doi.org/10.3390/publications5020009

Coulter, A. (2012). Patient Engagement—What Works? *The Journal of Ambulatory Care Management*, *35*(2). https://journals.lww.com/ambulatorycaremanagement/Fulltext/2012/04000/Patient_Engagement_What_Works_.3.aspx

Creswell, J. W., & Clark, V. L. P. (2017). *Designing and conducting mixed methods research*. Sage publications.

Crocker, J. C., Boylan, A.-M., Bostock, J., & Locock, L. (2017). Is it worth it? Patient and public views on the impact of their involvement in health research and its assessment: A UK-based qualitative interview study. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *20*(3), 519–528. https://doi.org/10.1111/hex.12479

Crockett, L. K., Shimmin, C., Wittmeier, K. D. M., & Sibley, K. M. (2019). Engaging patients and the public in Health Research: Experiences, perceptions and training needs among Manitoba health researchers. *Research Involvement and Engagement*, *5*(1), 28. https://doi.org/10.1186/s40900-019-0162-2

Crowe, S., Cresswell, K., Robertson, A., Huby, G., Avery, A., & Sheikh, A. (2011a). The case study approach. *BMC Medical Research Methodology*, *11*(1), 100. https://doi.org/10.1186/1471-2288-11-100

Crowe, S., Cresswell, K., Robertson, A., Huby, G., Avery, A., & Sheikh, A. (2011b). The case study approach. *BMC Medical Research Methodology*, *11*(1), 100. https://doi.org/10.1186/1471-2288-11-100

Culyer, A. J. (2016). HTA - Algorithm or Process? Comment on “Expanded HTA: Enhancing Fairness and Legitimacy”. *International Journal of Health Policy and Management*, *5*(8), 501–505. https://doi.org/10.15171/ijhpm.2016.59

Culyer, A. J., & Bombard, Y. (2012). *An Equity Framework for Health Technology Assessments*. *June*, 428–441. https://doi.org/10.1177/0272989X11426484

Davis, S. F., Silvester, A., Barnett, D., Farndon, L., & Ismail, M. (2019). Hearing the voices of older adult patients: Processes and findings to inform health services research. *Research Involvement and Engagement*, *5*(1), 11. https://doi.org/10.1186/s40900-019-0143-5

Dawson, S., Campbell, S. M., Giles, S. J., Morris, R. L., & Cheraghi-Sohi, S. (2018). Black and minority ethnic group involvement in health and social care research: A systematic review. *Health Expectations*, *21*(1), 3–22. https://doi.org/10.1111/hex.12597

De Santis, M., Hervas, C., Weinman, A., Bosi, G., & Bottarelli, V. (2019). Patient empowerment of people living with rare diseases. Its contribution to sustainable and resilient healthcare systems. *Annali Dell'Istituto Superiore Di Sanita*, *55*(3), Article 3. https://doi.org/10.4415/ANN_19_03_15

de Wit, M., Guillemin, F., Grimm, S., Boonen, A., Fautrel, B., & Joore, M. (2020). Patient engagement in health technology assessment (HTA) and the regulatory process: What about rheumatology?. *RMD Open*, *6*(3). https://doi.org/10.1136/rmdopen-2020-001286

Dewidar, O., Tsang, P., León-García, M., Mathew, C., Antequera, A., Baldeh, T., Akl, E. A., Alonso-Coello, P., Petkovic, J., Piggott, T., Pottie, K., Schünemann, H., Tugwell, P., & Welch, V. (2020). Over half of the WHO guidelines published from 2014 to 2019 explicitly considered health equity issues: A cross-sectional survey. *Journal of Clinical Epidemiology*, *127*, 125–133. https://doi.org/10.1016/j.jclinepi.2020.07.012

Dipankui, M. T., Gagnon, M.-P., Desmartis, M., Légaré, F., Piron, F., Gagnon, J., Rhiands, M., & Coulombe, M. (2015). Evaluation of Patient Involvement in a Health Technology Assessment. *International Journal of Technology Assessment in Health Care*, *31*(03), 166–170. https://doi.org/10.1017/S0266462315000240

Domecq, J. P., Prutsky, G., Elraiyah, T., Wang, Z., Nabhan, M., Shippee, N., Brito, J. P., Boehmer, K., Hasan, R., Firwana, B., Erwin, P., Eton, D., Sloan, J., Montori, V., Asi, N., Dabrh, A. M. A., & Murad, M. H. (2014). Patient engagement in research: A systematic review. *BMC Health Services Research*, *14*, 1–9. https://doi.org/10.1016/j.transproceed.2016.08.016

Douglas, C. M. W., Wilcox, E., Burgess, M., & Lynd, L. D. (2015). Why orphan drug coverage reimbursement decision-making needs patient and public involvement. *Health Policy (Amsterdam, Netherlands)*, *119*(5), 588–596. https://doi.org/10.1016/j.healthpol.2015.01.009

Doupi, P. (2016). Evolving Health IT Systems Evaluation: The Convergence of Health Informatics and HTA. *Studies in Health Technology and Informatics*, *222*, 220–236. http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med12&NEWS=N&AN=27198105

Dukhanin, V., Feeser, S., Berkowitz, S. A., & DeCamp, M. (2020). Who represents me? A patient-derived model of patient engagement via patient and family advisory councils (PFACs). *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *23*(1), 148–158. https://doi.org/10.1111/hex.12983

Eccles, A., Bryce, C., Turk, A., & Atherton, H. (2018). Patient and public involvement mobile workshops – convenient involvement for the un-usual suspects. *Research Involvement and Engagement*, *4*(1), 38. https://doi.org/10.1186/s40900-018-0123-1

Edgman-Levitan, S., & Schoenbaum, S. C. (2021). Patient-centered care: Achieving higher quality by designing care through the patient's eyes. *Israel Journal of Health Policy Research*, *10*(1), 21. https://doi.org/10.1186/s13584-021-00459-9

Ellard-Gray, A., Jeffrey, N. K., Choubak, M., & Crann, S. E. (2015). Finding the Hidden Participant: Solutions for Recruiting Hidden, Hard-to-Reach, and Vulnerable Populations. *International Journal of Qualitative Methods*, *14*(5), 1609406915621420. https://doi.org/10.1177/1609406915621420

Eslava-Schmalbach, J., Mosquera, P., Alzate, J. P., Pottie, K., Welch, V., Akl, E. A., Jull, J., Lang, E., Katikireddi, S. V., Morton, R., Thabane, L., Shea, B., Stein, A. T., Singh, J., Florez, I. D., Guyatt, G., Schunemann, H., & Tugwell, P. (2017). Considering health equity when moving from evidence-based guideline recommendations to implementation:

A case study from an upper-middle income country on the GRADE approach. *Health Policy and Planning*, *32*(10), 1484–1490. https://doi.org/10.1093/heapol/czx126

Espinoza, M. A., & Cabieses, B. (2014). [Equity in health and health technology assessment in Chile]. *Revista Medica de Chile, 142 Suppl*, S45-9. https://doi.org/10.4067/S0034-98872014001300008

Facey Helle, K. M., Hansen, P., Single, A. N. V., European Patients' Forum, Facey Helle, K. M., Hansen, P., & Single, A. N. V. (2017). *Patient Involvement in Health Technology Assessment* (Springer N, Vol. 32, Issue 2, p. 49). Springer Singapore. https://doi.org/10.1007/978-981-10-4068-9

Facey, K. M. (2019). As health technology assessment evolves so must its approach to patient involvement. *Journal of Comparative Effectiveness Research*, *8*(8), 549–554. https://doi.org/10.2217/cer-2019-0039

Facey, K. M., Bedlington, N., Berglas, S., Bertelsen, N., Single, A. N. V., & Thomas, V. (2018). Putting Patients at the Centre of Healthcare: Progress and Challenges for Health Technology Assessments. *The Patient*, *11*(6), 581–589. https://doi.org/10.1007/s40271-018-0325-5

Finset, A. (2017). Patient Participation, Engagement and Activation: Increased emphasis on the role of patients in healthcare. *Patient Education and Counseling*, *100*(7), 1245–1246. https://doi.org/10.1016/j.pec.2017.05.011

Fischer, J., & Van de Bovenkamp, H. M. (2019). The challenge of democratic patient representation: Understanding the representation work of patient organizations through methodological triangulation. *Health Policy*, *123*(1), 109–114. https://doi.org/10.1016/j.healthpol.2018.11.011

Fisher, I., & Ziviani, J. (2004). Explanatory case studies: Implications and applications for clinical research. *Australian Occupational Therapy Journal*, *51*(4), 185–191. https://doi.org/10.1111/j.1440-1630.2004.00446.x

Flanagin, A., Frey, T., Christiansen, S. L., & AMA Manual of Style Committee. (2021). Updated Guidance on the Reporting of Race and Ethnicity in Medical and Science Journals. *JAMA*, *326*(7), 621–627. https://doi.org/10.1001/jama.2021.13304

Forsythe, L. P., Carman, K. L., Szydlowski, V., Fayish, L., Davidson, L., Hickam, D. H., Hall, C., Bhat, G., Neu, D., Stewart, L., Jalowsky, M., Aronson, N., & Anyanwu, C. U. (2019). Patient Engagement In Research: Early Findings From The Patient-Centered Outcomes Research Institute. *Health Affairs*, *38*(3), 359–367. https://doi.org/10.1377/hlthaff.2018.05067

Frank, L., Morton, S. C., Guise, J.-M., Jull, J., Concannon, T. W., Tugwell, P., & Multi Stakeholder Engagement (MuSE) Consortium. (2020). Engaging Patients and Other Non-Researchers in Health Research: Defining Research Engagement. *Journal of General Internal Medicine*, *35*(1), 307–314. https://doi.org/10.1007/s11606-019-05436-2

Frohlich, K. L., & Potvin, L. (2008). Transcending the Known in Public Health Practice The Inequality Paradox: The Population Approach and Vulnerable Populations. *American Journal of Public Health*, *98*(2). https://doi.org/10.2105/AJPH.2007.114777

Gagné, T., & Veenstra, G. (2017). Inequalities in Hypertension and Diabetes in Canada: Intersections between Racial Identity, Gender, and Income. *Ethnicity & Disease*, *27*(4), 371–378. PubMed. https://doi.org/10.18865/ed.27.4.371

Gill, G. K., McNally, M. J., & Berman, V. (2018). Effective diversity, equity, and inclusion practices. *Healthcare Management Forum, 31*(5), 196–199. https://doi.org/10.1177/0840470418773785

Giuntella, O., Hyde, K., Saccardo, S., & Sadoff, S. (2021). Lifestyle and mental health disruptions during COVID-19. *Proceedings of the National Academy of Sciences of the United States of America*, *118*(9). https://doi.org/10.1073/pnas.2016632118

Goodman, M. S., & Sanders Thompson, V. L. (2017). The science of stakeholder engagement in research: Classification, implementation, and evaluation. *Translational Behavioral Medicine*, *7*(3), 486–491. https://doi.org/10.1007/s13142-017-0495-z

Graham, I. D., Logan, J., Harrison, M. B., Straus, S. E., Tetroe, J., Caswell, W., & Robinson, N. (2006). Lost in knowledge translation: Time for a map? *Journal of Continuing Education in the Health Professions*, *26*(1). https://journals.lww.com/jcehp/Fulltext/2006/26010/Lost_in_knowledge_translation__Time_for_a_map_.3.aspx

Grande, S. W., Faber, M. J., Durand, M. A., Thompson, R., & Elwyn, G. (2014). A classification model of patient engagement methods and assessment of their feasibility in real-world settings. *Patient Education and Counseling*, *95*(2), 281–287. https://doi.org/10.1016/j.pec.2014.01.016

Greenhalgh, T., Hinton, L., Finlay, T., Macfarlane, A., Fahy, N., Clyde, B., & Chant, A. (2019). Frameworks for supporting patient and public involvement in research: Systematic review and co-design pilot. *Health Expectations*, *22*(4), 785–801. https://doi.org/10.1111/hex.12888

Guindo, L. A., Wagner, M., Baltussen, R., Rindress, D., van Til, J., Kind, P., & Goetghebeur, M. M. (2012). From efficacy to equity: Literature review of decision criteria for resource allocation and healthcare decisionmaking. *Cost Effectiveness and Resource Allocation*. https://doi.org/10.1186/1478-7547-10-9

Gunn, C. J., Bertelsen, N., Regeer, B. J., & Schuitmaker-Warnaar, T. J. (2021). Valuing patient engagement: Reflexive learning in evidence generation practices for health technology assessment. *Social Science & Medicine (1982)*, *280*(ut9, 8303205), 114048. https://doi.org/10.1016/j.socscimed.2021.114048

Gutiérrez-Ibarluzea, I., Chiumente, M., & Dauben, H.-P. (2017). The Life Cycle of Health Technologies. Challenges and Ways Forward. *Frontiers in Pharmacology*, *8*, 14. https://doi.org/10.3389/fphar.2017.00014

Haerry, D., Landgraf, C., Warner, K., Hunter, A., Klingmann, I., May, M., & See, W. (2018). EUPATI and Patients in Medicines Research and Development: Guidance for Patient Involvement in Regulatory Processes. *Frontiers in Medicine*, *5*, 230–230. https://doi.org/10.3389/fmed.2018.00230

Hahn, D. L., Hoffmann, A. E., Felzien, M., LeMaster, J. W., Xu, J., & Fagnan, L. J. (2017). Tokenism in patient engagement. *Family Practice*, *34*(3). https://doi.org/10.1093/fampra/cmw097

Hanney, S. R., Grant, J., Wooding, S., & Buxton, M. J. (2004). Proposed methods for reviewing the outcomes of health research: The impact of funding by the UK's `Arthritis Research Campaign'. *Health Res Policy Syst*, *2*. https://doi.org/10.1186/1478-4505-2-4

Hashem, F., Calnan, M. W., & Brown, P. R. (2018). Decision making in NICE single technological appraisals: How does NICE incorporate patient perspectives?. *Health*

Expectations : An International Journal of Public Participation in Health Care and Health Policy, *21*(1), 128–137. https://doi.org/10.1111/hex.12594

Hicks, N., Hawken, N. A., Arvin-Berod, C., & Toumi, M. (2014). A Literature Review of Patient Advocacy Group (Pag) Involvement in HTA. *Value in Health : The Journal of the International Society for Pharmacoeconomics and Outcomes Research*, *17*(7), A438–A438. https://doi.org/10.1016/j.jval.2014.08.1139

Higgins, T., Larson, E., & Schnall, R. (2017). Unraveling the meaning of patient engagement: A concept analysis. *Patient Education and Counseling*, *100*(1), 30–36. https://doi.org/10.1016/j.pec.2016.09.002

Hill, S. R., & Olson, L. G. (2014). NICE, social values, and balancing objectivity and equity. *PharmacoEconomics*, *32*(11), 1039–1041. https://doi.org/10.1007/s40273-014-0220-4

Hivon, M., Lehoux, P., Denis, J.-L., & Tailliez, S. (2005). Use of health technology assessment in decision making: Coresponsibility of users and producers? *International Journal of Technology Assessment in Health Care*, *21*(2), 268–275. https://doi.org/10.1017/S0266462305050361

Hoch, J. S., Trenaman, L., Hearney, S. M., & Dewa, C. S. (2021). How Economic Decision Modeling Can Facilitate Health Equity. *AMA Journal of Ethics*, *23*(8), E624-630. https://doi.org/10.1001/amajethics.2021.624

Hosking, J., Macmillan, A., Jones, R., Ameratunga, S., & Woodward, A. (2019). Searching for health equity: Validation of a search filter for ethnic and socioeconomic inequalities in transport. *Systematic Reviews*, *8*(1), 94. https://doi.org/10.1186/s13643-019-1009-5

Huber, M., André Knottnerus, J., Green, L., Van Der Horst, H., Jadad, A. R., Kromhout, D., Leonard, B., Lorig, K., Loureiro, M. I., Van Der Meer, J. W. M., Schnabel, P., Smith, R., Van Weel, C., & Smid, H. (2011). How should we define health? *BMJ (Online)*, *343*(7817), 1–3. https://doi.org/10.1136/bmj.d4163

Huls, S. P. I., Whichello, C. L., van Exel, J., Uyl-de Groot, C. A., & de Bekker-Grob, E. W. (2019). What Is Next for Patient Preferences in Health Technology Assessment? A Systematic Review of the Challenges. *Value in Health : The Journal of the International Society for Pharmacoeconomics and Outcomes Research*, *22*(11), 1318–1328. https://doi.org/10.1016/j.jval.2019.04.1930

Jacklin, K., & Kinoshameg, P. (2008). Developing a Participatory Aboriginal Health Research Project: "Only if it's Going to Mean Something." *Journal of Empirical Research on Human Research Ethics*, *3*, 53–67.

Jackson, C. B., Quetsch, L. B., Brabson, L. A., & Herschell, A. D. (2018). Web-Based Training Methods for Behavioral Health Providers: A Systematic Review. *Administration and Policy in Mental Health*, *45*(4), 587–610. PubMed. https://doi.org/10.1007/s10488-018-0847-0

Jenkins, D. G., & Quintana-Ascencio, P. F. (2020). A solution to minimum sample size for regressions. *PloS One*, *15*(2), e0229345–e0229345. PubMed. https://doi.org/10.1371/journal.pone.0229345

Jenssen, B. P., Mitra, N., Shah, A., Wan, F., & Grande, D. (2016). Using Digital Technology to Engage and Communicate with Patients: A Survey of Patient Attitudes. *Journal of General Internal Medicine*, *31*(1), 85–92. https://doi.org/10.1007/s11606-015-3517-x

Johri, M., & OF, N. (2012). Can cost-effectiveness analysis integrate concerns for equity? Systematic review. *International Journal of Technology Assessment in Health Care*, *28*(2), 125–132. https://doi.org/10.1017/S0266462312000050

Kaushik, V., & Walsh, C. A. (2019). Pragmatism as a Research Paradigm and Its Implications for Social Work Research. *Social Sciences*, *8*(9). https://doi.org/10.3390/socsci8090255

Kent, S. M., & Yellowlees, P. (2015). The Technology-Enabled Patient Advocate: A Valuable Emerging Healthcare Partner. *Telemedicine and E-Health*, *21*(12), 1030–1037. https://doi.org/10.1089/tmj.2015.0025

Khalili, M., Karamouzian, M., Nasiri, N., Javadi, S., Mirzazadeh, A., & Sharifi, H. (2020). Epidemiological characteristics of COVID-19: A systematic review and meta-analysis. *Epidemiology and Infection*, *148*, e130–e130. PubMed. https://doi.org/10.1017/S0950268820001430

Khuntia, J., Ning, X., Cascio, W., & Stacey, R. (2022). Valuing Diversity and Inclusion in Health Care to Equip the Workforce: Survey Study and Pathway Analysis. *JMIR Formative Research*, *6*(5), e34808–e34808. PubMed. https://doi.org/10.2196/34808

Kingod, N., Cleal, B., Wahlberg, A., & Husted, G. R. (2016). Online Peer-to-Peer Communities in the Daily Lives of People With Chronic Illness: A Qualitative Systematic Review. *Qualitative Health Research*, *27*(1), 89–99. https://doi.org/10.1177/1049732316680203

Knaapen, L., & Lehoux, P. (2016). Three Conceptual Models of Patient and Public Involvement in Standard-setting: From Abstract Principles to Complex Practice. *Science as Culture*, *25*(2), 239–263. https://doi.org/10.1080/09505431.2015.1125875

Korchagina, D., Jaroslawski, S., Jadot, G., & Toumi, M. (2019). Orphan Drugs in Oncology. *Recent Results in Cancer Research. Fortschritte Der Krebsforschung. Progres Dans Les Recherches Sur Le Cancer*, *213*, 109–142. https://doi.org/10.1007/978-3-030-01207-6_8

Krausz, M., Westenberg, J. N., Vigo, D., Spence, R. T., & Ramsey, D. (2020). Emergency Response to COVID-19 in Canada: Platform Development and Implementation for eHealth in Crisis Management. *JMIR Public Health and Surveillance*, *6*(2), e18995. https://doi.org/10.2196/18995

Krieger, N. (2014). Discrimination and health inequities. *International Journal of Health Services : Planning, Administration, Evaluation*, *44*(4), 643–710. https://doi.org/10.2190/HS.44.4.b

Lambert, R., Carter, D., Burgess, N., & Haji Ali Afzali, H. (2018). The development of funding recommendations for health technologies at the state level: A South Australian case study. *The International Journal of Health Planning and Management*, *33*(4), 806–822. https://doi.org/10.1002/hpm.2529

Lawn, S., Zhi, X., & Morello, A. (2017). An integrative review of e-learning in the delivery of self-management support training for health professionals. *BMC Medical Education*, *17*(1), 183. https://doi.org/10.1186/s12909-017-1022-0

Liddy, C., Hunter, Z., Mihan, A., & Keely, E. (2017). Use of Facebook as part of a social media strategy for patient engagement. *Canadian Family Physician Medecin de Famille Canadien*, *63*(3), 251–252. PubMed. https://pubmed.ncbi.nlm.nih.gov/28292805

Lopes, E., Street, J., Carter, D., & Merlin, T. (2016). Involving patients in health technology funding decisions: Stakeholder perspectives on processes used in Australia. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *19*(2), 331–344. https://doi.org/10.1111/hex.12356

Macklin, R. (2010). Enrolling pregnant women in biomedical research. *The Lancet*, *375*(9715), 632–633. https://doi.org/10.1016/S0140-6736(10)60257-7

MacPhail, E., & Shea, B. (2017). *An Inside Look at the Early History of the CADTH Common Drug Review in Canada* (p. 68). Canadian Agency for Drugs and Technologies in Health. https://www.cadth.ca/sites/default/files/pdf/early_history_of_CDR.pdf

Maguire, K., & Britten, N. (2017). "How can anybody be representative for those kind of people?" Forms of patient representation in health research, and why it is always contestable. *Social Science and Medicine*, *183*, 62–69. https://doi.org/10.1016/j.socscimed.2017.04.049

Maher, C., Hadfield, M., Hutchings, M., & de Eyto, A. (2018). Ensuring Rigor in Qualitative Data Analysis: A Design Research Approach to Coding Combining NVivo With Traditional Material Methods. *International Journal of Qualitative Methods*, *17*(1), 1609406918786362. https://doi.org/10.1177/1609406918786362

Majid, U., & Gagliardi, A. (2019). Clarifying the degrees, modes, and muddles of "meaningful" patient engagement in health services planning and designing. *Patient Education and Counseling*, *102*(9), 1581–1589. https://doi.org/10.1016/j.pec.2019.04.006

Malterud, K., Siersma, V. D., & Guassora, A. D. (2016). Sample Size in Qualitative Interview Studies: Guided by Information Power. *Qualitative Health Research*, *26*(13), 1753–1760. https://doi.org/10.1177/1049732315617444

Manafo, E., Petermann, L., Mason-Lai, P., & Vandall-Walker, V. (2018). Patient engagement in Canada: A scoping review of the "how" and "what" of patient engagement in health research. *Health Research Policy and Systems*, *16*(1), 5–5. https://doi.org/10.1186/s12961-018-0282-4

Marmot, M. (2017). The Health Gap: The Challenge of an Unequal World: The argument. *International Journal of Epidemiology*, *46*(4), 1312–1318. PubMed. https://doi.org/10.1093/ije/dyx163

Marsh, K., & Le, A. (2015). *More Care Needed: Incorporating Equity into Health Technology Assessment*. *October*, 9–13.

Martin, D., Miller, A. P., Quesnel-Vallée, A., Caron, N. R., Vissandjée, B., & Marchildon, G. P. (2018). Canada's universal health-care system: Achieving its potential. *The Lancet*, *391*(10131), 1718–1735. https://doi.org/10.1016/S0140-6736(18)30181-8

Mazanderani, F., O'Neill, B., & Powell, J. (2013). "People power" or "pester power"? YouTube as a forum for the generation of evidence and patient advocacy. *Patient Education and Counseling*, *93*(3), 420–425. https://doi.org/10.1016/J.PEC.2013.06.006

Mazor, K. M., Rubin, D. L., Roblin, D. W., Williams, A. E., Han, P. K. J., Gaglio, B., Cutrona, S. L., Costanza, M. E., & Wagner, J. L. (2016). Health literacy-listening skill and patient questions following cancer prevention and screening discussions. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *19*(4), Article 4. https://doi.org/10.1111/hex.12387

Mbuagbaw, L., Aves, T., Shea, B., Jull, J., Welch, V., Taljaard, M., Yoganathan, M., Greer-Smith, R., Wells, G., & Tugwell, P. (2017). Considerations and guidance in designing equity-relevant clinical trials. *International Journal for Equity in Health*, *16*(1), Article 1. https://doi.org/10.1186/s12939-017-0591-1

McCoy, M. S., Warsh, J., Rand, L., Parker, M., & Sheehan, M. (2019). Patient and public involvement: Two sides of the same coin or different coins altogether? *Bioethics*, *33*(6), 708–715. https://doi.org/10.1111/bioe.12584

Melsen, W. G., Bootsma, M. C. J., Rovers, M. M., & Bonten, M. J. M. (2014). The effects of clinical and statistical heterogeneity on the predictive values of results from meta-analyses. *Clinical Microbiology and Infection*, *20*(2), 123–129. https://doi.org/10.1111/1469-0691.12494

Menon, D. (2015). Health technology assessment: The journey continues. *Canadian Medical Association Journal*, *187*(1), E19. https://doi.org/10.1503/cmaj.140698

Mercer, R. E., Chambers, A., Mai, H., McDonald, V., McMahon, C., & Chan, K. K. W. (2020). Are We Making a Difference? A Qualitative Study of Patient Engagement at the pan-Canadian Oncology Drug Review: Perspectives of Patient Groups. *Value in Health : The Journal of the International Society for Pharmacoeconomics and Outcomes Research*, *23*(9), 1157–1162. https://doi.org/10.1016/j.jval.2020.06.003

Misselbrook, D. (2014). W is for wellbeing and the WHO definition of health. *The British Journal of General Practice : The Journal of the Royal College of General Practitioners*, *64*(628), 582–582. https://doi.org/10.3399/bjgp14X682381

Mitchell, M., & Kan, L. (2019). Digital Technology and the Future of Health Systems. *Health Systems and Reform*, *5*(2), 113–120. https://doi.org/10.1080/23288604.2019.1583040

Mohara, A., Youngkong, S., Velasco, R. P., Werayingyong, P., Pachanee, K., Prakongsai, P., Tantivess, S., Tangcharoensathien, V., Lertiendumrong, J., Jongudomsuk, P., & Teerawattananon, Y. (2012). Using health technology assessment for informing coverage decisions in Thailand. *Journal of Comparative Effectiveness Research*, *1*(2), 137–146. https://doi.org/10.2217/cer.12.10

Moreira, T. (2015). Understanding the role of patient organizations in health technology assessment. *Health Expectations : An International Journal of Public Participation in Health Care and Health Policy*, *18*(6), 3349–3357. https://doi.org/10.1111/hex.12325

Muhlbacher, A. C., & Johnson, F. R. (2017). Giving Patients a Meaningful Voice in European Health Technology Assessments: The Role of Health Preference Research. *The Patient*, *10*(4), 527–530. https://doi.org/10.1007/s40271-017-0249-5

Ndaïrou, F., Area, I., Nieto, J. J., & Torres, D. F. M. (2020). Mathematical modeling of COVID-19 transmission dynamics with a case study of Wuhan. *Chaos, Solitons, and Fractals*, *135*, 109846–109846. PubMed. https://doi.org/10.1016/j.chaos.2020.109846

Nelson, J. (2017). Using conceptual depth criteria: Addressing the challenge of reaching saturation in qualitative research. *Qualitative Research*, *17*(5), 554–570. https://doi.org/10.1177/1468794116679873

Nowell, L. S., Norris, J. M., White, D. E., & Moules, N. J. (2017). Thematic Analysis: Striving to Meet the Trustworthiness Criteria. *International Journal of Qualitative Methods*, *16*(1), 1609406917733847. https://doi.org/10.1177/1609406917733847

Nyblade, L., Stockton, M. A., Giger, K., Bond, V., Ekstrand, M. L., Lean, R. M., Mitchell, E. M. H., Nelson, L. R. E., Sapag, J. C., Siraprapasiri, T., Turan, J., & Wouters, E. (2019). Stigma in health facilities: Why it matters and how we can change it. *BMC Medicine*, *17*(1), 25–25. PubMed. https://doi.org/10.1186/s12916-019-1256-2

Ocloo, J., & Matthews, R. (2016). From tokenism to empowerment: Progressing patient and public involvement in healthcare improvement. *BMJ Quality & Safety*, *25*(8), 626. https://doi.org/10.1136/bmjqs-2015-004839

O’Flaherty, M., Lloyd-Williams, F., Capewell, S., Boland, A., Maden, M., Collins, B., Bandosz, P., Hyseni, L., & Kypridemos, C. (2021). Modelling tool to support decision-making in the NHS Health Check programme: Workshops, systematic review and co-production with users. *Health Technology Assessment (Winchester, England)*, *25*(35), 1–234. https://doi.org/10.3310/hta25350

Ohannessian, R., Duong, T. A., & Odone, A. (2020). Global Telemedicine Implementation and Integration Within Health Systems to Fight the COVID-19 Pandemic: A Call to Action. *JMIR Public Health Surveill*, *6*(2), e18810. https://doi.org/10.2196/18810

O’Neill, J., Tabish, H., Welch, V., Petticrew, M., Pottie, K., Clarke, M., Evans, T., Pardo Pardo, J., Waters, E., White, H., & 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. https://doi.org/10.1016/j.jclinepi.2013.08.005

O’Rourke, B., Oortwijn, W., & Schuller, T. (2020). The new definition of health technology assessment: A milestone in international collaboration. *International Journal of Technology Assessment in Health Care*, *36*(3), 187–190. Cambridge Core. https://doi.org/10.1017/S0266462320000215

Pasternack, I., Anttila, H., Mäkelä, M., Ikonen, T., Räsänen, P., Lampe, K., Van Brabandt, H., Cleemput, I., De Laet, C., Marchetti, M., Refolo, P., Sacchini, D., Kunz, R., Nordmann, A., Lühmann, D., & Freyschuss, B. (2009). Testing the HTA core model: Experiences from two pilot projects. *International Journal of Technology Assessment in Health Care*. https://doi.org/10.1017/S026646230999064X

Patient Engagement Action Team. (2017). *Engaging patients in patient safety: A Canadian guide*. https://www.hqontario.ca/Portals/0/documents/pe/engaging-patients-in-patient-safety-en.pdf

Petkovic, J., Duench, S., Trawin, J., Dewidar, O., Pardo Pardo, J., Simeon, R., DesMeules, M., Gagnon, D., Hatcher Roberts, J., Hossain, A., Pottie, K., Rader, T., Tugwell, P., Yoganathan, M., Presseau, J., & Welch, V. (2021a). Behavioural interventions delivered through interactive social media for health behaviour change, health outcomes, and health equity in the adult population. *The Cochrane Database of Systematic Reviews*, *5*(5), Article 5. https://doi.org/10.1002/14651858.CD012932.pub2

Petkovic, J., Duench, S., Trawin, J., Dewidar, O., Pardo Pardo, J., Simeon, R., DesMeules, M., Gagnon, D., Hatcher Roberts, J., Hossain, A., Pottie, K., Rader, T., Tugwell, P., Yoganathan, M., Presseau, J., & Welch, V. (2021b). Behavioural interventions delivered through interactive social media for health behaviour change, health outcomes, and health equity in the adult population. *The Cochrane Database of Systematic Reviews*, *5*(5), CD012932. https://doi.org/10.1002/14651858.CD012932.pub2

Pinheiro, J., Bates, D., & R Core Team. (2022). *R package:Linear and Nonlinear Mixed Effects Models.* (Version 3.1-160. CRAN - Package nlme 2) [Computer software].

Prady, S. L., Uphoff, E. P., Power, M., & Golder, S. (2018). Development and validation of a search filter to identify equity-focused studies: Reducing the number needed to screen. *BMC Medical Research Methodology*, *18*(1), 106. https://doi.org/10.1186/s12874-018-0567-x

Press, Z., & Richards, D. (2015). The power of patient ownership: The path from engagement to equity. *Patient Experience Journal*, *2*(1), 15–17. §

Ramel, V. (2019). Addressing equity in eHealth interventions for healthcare system users & patients, literature review. *European Journal of Public Health*, *29*(Supplement_4), ckz186.091. https://doi.org/10.1093/eurpub/ckz186.091

Regnault, A., Willgoss, T., Barbic, S., & On behalf of the International Society for Quality of Life Research (ISOQOL) Mixed Methods Special Interest Group (SIG). (2018). Towards the use of mixed methods inquiry as best practice in health outcomes research. *Journal of Patient-Reported Outcomes*, *2*(1), 19. https://doi.org/10.1186/s41687-018-0043-8

Reynolds, J., Ogden, M., & Beresford, R. (2021). Conceptualising and constructing ‘diversity’ through experiences of public and patient involvement in health research. *Research Involvement and Engagement*, *7*(1), 53. https://doi.org/10.1186/s40900-021-00296-9

Richard, L., Furler, J., Densley, K., Haggerty, J., Russell, G., Levesque, J. F., Gunn, J., & Richard Lauralie and Furler, J. and D. K. and H. J. and R. G. and L. J.-F. and G. J. (2016). Equity of access to primary healthcare for vulnerable populations: The IMPACT international online survey of innovations. *International Journal for Equity in Health*, *15*(1), 64–64. https://doi.org/10.1186/s12939-016-0351-7

Roche, P., Shimmin, C., Hickes, S., Khan, M., Sherzoi, O., Wicklund, E., Lavoie, J. G., Hardie, S., Wittmeier, K. D. M., & Sibley, K. M. (2020). Valuing All Voices: Refining a trauma-informed, intersectional and critical reflexive framework for patient engagement in health research using a qualitative descriptive approach. *Research Involvement and Engagement*, *6*(1), 42. https://doi.org/10.1186/s40900-020-00217-2

Rowland, P., & Kumagai, A. K. (2018). Dilemmas of Representation: Patient Engagement in Health Professions Education. *Academic Medicine*, *93*(6).

https://journals.lww.com/academicmedicine/Fulltext/2018/06000/Dilemmas_of_Representation__Patient_Engagement_in.30.aspx

Rycroft-Malone, J., Burton, C. R., Bucknall, T., Graham, I. D., Hutchinson, A. M., & Stacey, D. (2016). Collaboration and Co-Production of Knowledge in Healthcare: Opportunities and Challenges. *International Journal of Health Policy and Management*, *5*(4), 221–223. PubMed. https://doi.org/10.15171/ijhpm.2016.08

Safaei, J. (2015a). Deliberative democracy in health care: Current challenges and future prospects. *Journal of Healthcare Leadership*, *7*, 123–136. https://doi.org/10.2147/JHL.S70021

Safaei, J. (2015b). Deliberative democracy in health care: Current challenges and future prospects. *Journal of Healthcare Leadership*, *7*, 123–136. https://doi.org/10.2147/JHL.S70021

Schoonenboom, J., & Johnson, R. B. (2017). How to Construct a Mixed Methods Research Design. *Kolner Zeitschrift Fur Soziologie Und Sozialpsychologie*, *69*(Suppl 2), 107–131. https://doi.org/10.1007/s11577-017-0454-1

Scott, A. M., Wale, J. L., & HTAi Patient and Citizen Involvement in HTA Interest Group, P. I. and E. W. G. (2017). Patient advocate perspectives on involvement in HTA: an international snapshot. *Research Involvement and Engagement*, *3*(1), 2–2. https://doi.org/10.1186/s40900-016-0052-9

Shams Moattar, A., Asghari, F., Majdzadeh, R., A, S. M., Asghari, F., Majdzadeh, R., Shams Moattar, A., Asghari, F., Majdzadeh, R., A, S. M., Asghari, F., & Majdzadeh, R. (2016). Do ethical considerations influence any in HTA reports? A review of reports. *Medical*

Journal of the Islamic Republic of Iran, *30*, 362–362. http://www.epistemonikos.org/documents/1196d00f0fe0fea57ee8c340c6bc379110ec91db

Sharma, A., Harrington, R. A., McClellan, M. B., Turakhia, M. P., Eapen, Z. J., Steinhubl, S., Mault, J. R., Majmudar, M. D., Roessig, L., Chandross, K. J., Green, E. M., Patel, B., Hamer, A., Olgin, J., Rumsfeld, J. S., Roe, M. T., & Peterson, E. D. (2018). Using Digital Health Technology to Better Generate Evidence and Deliver Evidence-Based Care. *Journal of the American College of Cardiology*, *71*(23), 2680–2690. https://doi.org/10.1016/j.jacc.2018.03.523

Sharma, P., & Kaur, P. D. (2017). Effectiveness of web-based social sensing in health information dissemination—A review. *Telematics and Informatics*, *34*(1), 194–219. https://doi.org/10.1016/J.TELE.2016.04.012

Shimmin, C., Wittmeier, K. D. M., Lavoie, J. G., Wicklund, E. D., & Sibley, K. M. (2017). Moving towards a more inclusive patient and public involvement in health research paradigm: The incorporation of a trauma-informed intersectional analysis. *BMC Health Services Research*, *17*(1), 539–539. https://doi.org/10.1186/s12913-017-2463-1

Shivayogi, P. (2013). Vulnerable population and methods for their safeguard. *Perspectives in Clinical Research*, *4*(1), 53–57. PubMed. https://doi.org/10.4103/2229-3485.106389

Shorten, A., & Smith, J. (2017). Mixed methods research: Expanding the evidence base. *Evidence Based Nursing*, *20*(3), 74. https://doi.org/10.1136/eb-2017-102699

Snow, M. E., Tweedie, K., & Pederson, A. (2018). Heard and valued: The development of a model to meaningfully engage marginalized populations in health services planning. *BMC Health Services Research*, *18*(1), 181–181. https://doi.org/10.1186/s12913-018-2969-1

Soares, M. O., Walker, S., Palmer, S. J., & Sculpher, M. J. (2018). Establishing the Value of Diagnostic and Prognostic Tests in Health Technology Assessment. *Medical Decision Making*, *38*(4), 495–508. https://doi.org/10.1177/0272989X17749829

Staley, K. (2015). 'Is it worth doing?' Measuring the impact of patient and public involvement in research. *Research Involvement and Engagement*, *1*(1). https://doi.org/10.1186/s40900-015-0008-5

Staley, K., & Doherty, C. (2016). It's not evidence, it's insight: Bringing patients' perspectives into health technology appraisal at NICE. *Research Involvement and Engagement*, *2*(1), 4–4. https://doi.org/10.1186/s40900-016-0018-y

Stephanie A Kraft, Erin Rothwell, Seema K Shah, Devan M Duenas, Hannah Lewis, Kristin Muessig, Douglas J Opel, Katrina A B Goddard, & Benjamin S Wilfond. (2021). Demonstrating 'respect for persons' in clinical research: Findings from qualitative interviews with diverse genomics research participants. *Journal of Medical Ethics*, *47*(12), e8. https://doi.org/10.1136/medethics-2020-106440

Stevens, A., Milne, R., & Burls, A. (2003). Health technology assessment: History and demand. *Journal of Public Health Medicine*, *25*(2), 98–101. https://doi.org/10.1093/pubmed/fdg022

Straus, S. E., Tetroe, J., & Graham, I. D. (2013). *Knowledge Translation in Health Care: Moving from Evidence to Practice—CIHR* (2nd ed.). http://www.cihr-irsc.gc.ca/e/40618.html

Sullivan, S. D., Watkins, J., Sweet, B., & Ramsey, S. D. (2009). Health Technology Assessment in Health-Care Decisions in the United States. *Value in Health*, *12*(s2), S39–S44. https://doi.org/10.1111/j.1524-4733.2009.00557.x

Swan, M. (2009). Emerging patient-driven health care models: An examination of health social networks, consumer personalized medicine and quantified self-tracking. *International Journal of Environmental Research and Public Health*, *6*(2), 492–525. PubMed. https://doi.org/10.3390/ijerph6020492

Szumilas, M. (2010). Explaining odds ratios. *Journal of the Canadian Academy of Child and Adolescent Psychiatry = Journal de l'Academie Canadienne de Psychiatrie de l'enfant et de l'adolescent*, *19*(3), 227–229.

Taherdoost, H. (2018). A review of technology acceptance and adoption models and theories. *Procedia Manufacturing*, *22*, 960–967. https://doi.org/10.1016/J.PROMFG.2018.03.137

Tarricone, R., Petracca, F., Cucciniello, M., & Ciani, O. (2022). Recommendations for developing a lifecycle, multidimensional assessment framework for mobile medical apps. *Health Economics*, *bvq*, *9306780*. https://doi.org/10.1002/hec.4505

Thurber, M. C., Warner, C., Platt, L., Slaski, A., Gupta, R., & Miller, G. (2013). To promote adoption of household health technologies, think beyond health. *American Journal of Public Health*, *103*(10), 1736–1740. https://doi.org/10.2105/AJPH.2013.301367

Ting, D. S. W., Carin, L., Dzau, V., & Wong, T. Y. (2020). Digital technology and COVID-19. *Nature Medicine*, *26*(4), 459–461. https://doi.org/10.1038/s41591-020-0824-5

Tugwell, P., De Savigny, D., Hawker, G., & Robinson, V. (2006a). Applying clinical epidemiological methods to health equity: The equity effectiveness loop. *British Medical Journal*. https://doi.org/10.1136/bmj.332.7537.358

Tugwell, P., De Savigny, D., Hawker, G., & Robinson, V. (2006b). Applying clinical epidemiological methods to health equity: The equity effectiveness loop. *British Medical Journal*. https://doi.org/10.1136/bmj.332.7537.358

van der Waal, J., Daenekindt, S., & de Koster, W. (2017). Statistical challenges in modelling the health consequences of social mobility: The need for diagonal reference models. *International Journal of Public Health*, *62*(9), 1029–1037. https://doi.org/10.1007/s00038-017-1018-x

Vedel, I., Kaur, N., Hong, Q. N., El Sherif, R., Khanassov, V., Godard-Sebillotte, C., Sourial, N., Yang, X. Q., & Pluye, P. (2019). Why and how to use mixed methods in primary health care research. *Family Practice*, *36*(3), 365–368. PubMed. https://doi.org/10.1093/fampra/cmy127

Vreman, R. A., Mantel-Teeuwisse, A. K., Hövels, A. M., Leufkens, H. G. M., & Goettsch, W. G. (2020). Differences in Health Technology Assessment Recommendations Among European Jurisdictions: The Role of Practice Variations. *Value in Health*, *23*(1), 10–16. https://doi.org/10.1016/j.jval.2019.07.017

Wahlster, P., Brereton, L., Burns, J., Hofmann, B., Mozygemba, K., Oortwijn, W., Pfadenhauer, L., Polus, S., Rehfuess, E., Schilling, I., van der Wilt, G. J., & Gerhardus, A. (2017a). AN INTEGRATED PERSPECTIVE ON THE ASSESSMENT OF TECHNOLOGIES: INTEGRATE-HTA. *International Journal of Technology Assessment in Health Care*, *33*(5), 544–551. https://doi.org/10.1017/S0266462317000484

Wahlster, P., Brereton, L., Burns, J., Hofmann, B., Mozygemba, K., Oortwijn, W., Pfadenhauer, L., Polus, S., Rehfuess, E., Schilling, I., van der Wilt, G. J., & Gerhardus, A. (2017b). AN INTEGRATED PERSPECTIVE ON THE ASSESSMENT OF TECHNOLOGIES: INTEGRATE-HTA. *International Journal of Technology Assessment in Health Care*, *33*(5), Article 5. https://doi.org/10.1017/S0266462317000484

Wale, J. L., Scott, A. M., Bertelsen, N., Meade, N., & (PCIG), Hta. P. and C. I. in H. T. A. I. G. (2017). Strengthening international patient advocacy perspectives on patient involvement in HTA within the HTAi Patient and Citizen Involvement Interest Group—Commentary. *Research Involvement and Engagement*, *3*, 3–3. https://doi.org/10.1186/s40900-016-0053-8

Wale, J. L., Thomas, S., Hamerlijnck, D., & Hollander, R. (2021). Patients and public are important stakeholders in health technology assessment but the level of involvement is low – a call to action. *Research Involvement and Engagement*, *7*(1), 1. https://doi.org/10.1186/s40900-020-00248-9

Wale, J., Scott, A. M., Hofmann, B., Garner, S., Low, E., & Sansom, L. (2017). Why patients should be involved in health technology assessment. *International Journal of Technology Assessment in Health Care*, *33*(1), 1–4. https://doi.org/10.1017/S0266462317000241

Wang, Z., & Tang, K. (2020). Combating COVID-19: Health equity matters. *Nature Medicine, 26*(4), 458–458. https://doi.org/10.1038/s41591-020-0823-6

Ward, L. M., Hill, M. J., Picard, A., Olsen Harper, A., Chreim, S., & Wells, S. (2021). A process of healing for the Labrador Innu: Improving health and wellbeing in the context of historical and contemporary colonialism. *Social Science & Medicine*, *279*, 113973. https://doi.org/10.1016/j.socscimed.2021.113973

Warsh, J. (2014). PPI: Understanding the Difference Between Patient and Public Involvement. *The American Journal of Bioethics*, *14*(6), 25–26. https://doi.org/10.1080/15265161.2014.900144

Welch, V. A., Mosdol, A., Murad, M. H., Solà, I., Akl, E. A., Matovinovic, E., Vitols, S., Briel, M., Ansari, M. T., Shea, B., Eslava-Schmalbach, J., Guyatt, G., Sharaf, R., Katikireddi,

S. V., Jull, J., Watine, J., Dans, L., Morton, R. L., Singh, J. A., … Stanev, R. (2017a). GRADE equity guidelines 3: Considering health equity in GRADE guideline development: Rating the certainty of synthesized evidence. *Journal of Clinical Epidemiology, 90*(2017), 76–83. https://doi.org/10.1016/j.jclinepi.2017.01.015

Welch, V. A., Mosdol, A., Murad, M. H., Solà, I., Akl, E. A., Matovinovic, E., Vitols, S., Briel, M., Ansari, M. T., Shea, B., Eslava-Schmalbach, J., Guyatt, G., Sharaf, R., Katikireddi, S. V., Jull, J., Watine, J., Dans, L., Morton, R. L., Singh, J. A., … Stanev, R. (2017b). GRADE equity guidelines 3: Considering health equity in GRADE guideline development: Rating the certainty of synthesized evidence. *Journal of Clinical Epidemiology, 90*(2017), Article 2017. https://doi.org/10.1016/j.jclinepi.2017.01.015

Welch, V., Tugwell, P., & Morris, E. B. (2008). The Equity-effectiveness Loop as a Tool for Evaluating Population Health Interventions. *Revista de Salud Pública, 10*, 83–96. https://doi.org/10.1590/S0124-00642008000600008

Whitty, J. A., & Littlejohns, P. (2015). Social values and health priority setting in Australia: An analysis applied to the context of health technology assessment. *Health Policy (Amsterdam, Netherlands), 119*(2), 127–136. https://doi.org/10.1016/j.healthpol.2014.09.003

Wilkinson-Stokes, M., Kellermeier, M., & Whitfield, S. (2023). ‘Send everyone, it’s my son’ – Combined Glaserian grounded theory and thematic analysis of paramedics attending their own families. *Paramedicine*, *20*(5), 161–176. https://doi.org/10.1177/27536386231178961

Wilson, K., & Flood, C. M. (2021). Implementing digital passports for SARS-CoV-2 immunization in Canada. *Canadian Medical Association Journal*, *193*(14), E486. https://doi.org/10.1503/cmaj.210244

Wortley, S., Wale, J., Grainger, D., & Murphy, P. (2017). Moving beyond the rhetoric of patient input in health technology assessment deliberations. *Australian Health Review : A Publication of the Australian Hospital Association*, *41*(2), 170–172. https://doi.org/10.1071/AH15216

Yao, R., Zhang, W., Evans, R., Cao, G., Rui, T., & Shen, L. (2022). Inequities in Health Care Services Caused by the Adoption of Digital Health Technologies: Scoping Review. *Journal of Medical Internet Research*, *24*(3), e34144. https://doi.org/10.2196/34144

Appendices

Appendix A: Stakeholder input to inform thesis proposal (2017-2018)

Relevant Research questions	Current issues/ concerns
How vulnerable people are engaged in health-related initiatives how to improve patient groups 'expertise (HTA analysts)	Challenges to collect information from patients, not sure if data reported by patient group representatives are from patients; (HTA analyst)
How to provide support to patient groups for effective participation in HTA (Patient group) What are the best practices in Patient Engagement? (HTA analyst) - Does individual patient really feel engaged in HTA process? - What is the level of involvement of patients in HTA? - What influence patients have in HTA? - From a PG perspective, what are their frustrations?	- Current process is not very interactive - Feel disconnected sometime - There is no dynamic exchange in the current process - PGs are criticized for taking money from pharma industry, but current criteria do not provide impetus to apply for grant - Does not have resources to apply for grant - PG does not have control on the price of medication (Patient group)
How to build capacity for patient engagement? How to build health equity in patient engagement process? How to situate patient lived experiences in the HTA process? (HTA analyst)	It is hard to know whose voice is represented Digital divide: certain populations with vested interest in the outcomes of patient engagement cannot participate We have the researcher but not the diversity or patients; health literacy and access to vulnerable groups (immigrants etc. is an issue)
How to describe vulnerabilities for people who are non-POC. health equity and have rare diseases (Patients)	Patient groups are not funded; we struggle a lot to get our voices across. (Patient group)
General advice to involve patients in my research: - Provide some monetary incentive to patients on the advisory group and research participants (Patients)	

Appendix B: Data extraction form

Characteristics of reports						
Brand Name and weblink	Publication year(Recommendation issued)	HTA organizations	types of health technologies	Generic Name	Therapeutic Area/Indication	
Analysis questions : PROGRESS-PLUS , EQUITY CHECKLIST and Practical Guidance for Involving Stakeholders in Health Research						
Q1: What were the final recommendations?	Q2: Do the recommendations account for any aspects through which inequities can emerge when analysing the condition or disease? If yes, use PROGRESS-Plus to identify the population groups affected by those inequities-	Q3Are the recommendations generalizable to all population groups that are/might be affected by the treatment or services? i.e. was sub-group analysis conducted If yes, use PROGRESS-Plus to identify the population groups affected by those inputs	Q4: Are patient inputs/ preferences/ values/ reported in the HTA RECOMMENDATIONS ?	Q5: Are patient inputs/ preferences/ values generalizable to all population groups that are/might be affected by the treatment or condition/ was sub-group analysis conducted? If yes, use PROGRESS-Plus to identify the population groups affected by those items	Q6: Did the HTA organization involve patient organizations/ groups? if yes, record the name(s) of patient organizations/groups involved RECORD STATEMENTS IN SEPARATE COLUNMS	Q7: Did patients contribute to decision-making? (deliberative framework) - Did patient participate in formulating recommendation - patients 'role in deliberative framework (Primarily vote for non-cancer, consensus for cancer, consensus for HQO)
Q8:Did individual patients/patient group provide feedback or input?	Q9: Did the HTA organization involve individual patients/caregivers not affiliated with a patient group?	Q10: Who provided patients' perspectives?	Q11: Did the patients who provided inputs have experience with the condition or the treatment?	Q12: What methods do HTA organizations use to collect patients 'perspectives?	Q13 What methods do patient organizations/groups use to collect patients' perspectives?	14: Types of digital technologies used in reports

Appendix C: Consent forms

Consent Form – Patients

Title of the study: Patient engagement for equity-focused health technology assessment (HTA) recommendations in the digital era

Principal Investigator: Rosiane Simeon, MPH, PhD (c)
Interdisciplinary School of Health Sciences, Faculty of Health Sciences, University of Ottawa
Phone: email:

Supervisors: Dr. Vivian Welch: and Dr. Peter Tugwell

School of Epidemiology and Public Health, Faculty of Medicine, University of Ottawa

Invitation to Participate: I am invited to participate in the abovementioned research study conducted by Rosiane Simeon, a PhD candidate supervised by Dr. Vivian Welch and Dr. Peter Tugwell.

Purpose of the Study: The purpose of the study is to assess the use of digital technologies for patient engagement to promote the inclusion and participation of patients with diverse background for informing the development of HTA recommendations that are reasonable and reflect the preferences of patients.

Participation: My participation will consist essentially of doing an interview with the principal investigator- Rosiane Simeon. The interview will take approximately 45-60 minutes during which I will share my experiences and perspectives about the use of digital technologies for inclusive and diverse patient engagement to inform HTA recommendations. I will communicate with the principal investigator to find a suitable date and time for the interview, which will take place online via Zoom. The interview will be audio recorded only.

Risks: My participation in this study will entail that I share my name, my title and specific role(s), the name of my affiliated organization and information on my personal experiences about participating in patient engagement activities. The researcher does not anticipate any major risks associated with me sharing the above information. The information that I share about my experiences will not be associated with me and my name will not be included in the publication of the results; only the researcher will have access to this information.

Benefits: My participation in this study will help contribute to the advancement of knowledge for developing patient engagement strategies that are inclusive of patients from diverse background, and that consider patients' perspectives in HTA recommendations.

Confidentiality and anonymity: I have received assurance from the researcher that the information I will share will remain strictly confidential.

I understand that the content will be used only to identify factors for the use of digital technologies to support patient engagement strategies and that my confidentiality will be protected and that no one other than the researcher will have access to the recording.

Anonymity will be protected in the following manner: interviews will be audio recorded only each interview will be de-identified and labelled using a unique alpha-numeric. All interviews will be transcribed by the principal investigator.

Written quotations of comments will be used in publications but de-identified when possible.

I understand that it might be impossible to completely conceal the identity of patient organizations because it is a small community of research, where most organizations share knowledge and practices. Some organizations may be identifiable based on quotation content.

I therefore selected Yes or No in the following options:

1. [] Yes, the researcher can use my title(s) in quotes (e.g. coordinator)
2. [] No, the researcher cannot use my title(s) in quotes (e.g. patient 23B)
3. [] Yes, the researcher can link me to my organization (e.g. coordinator/patient of X organization)
4. [] No, the researcher cannot link me to my organization (e.g. coordinator of unnamed organization)

Conservation of data: The data collected recordings and transcripts of interviews will be kept in a secure manner using alpha-numeric passwords on the principal investigator's computer. All data will be stored at the principal investigator's home for 5 years and will be accessed only by the principal investigator.

Compensation: I will receive a $ 25 CAD gift card for completing the interview. It is a way for the principal investigator to show appreciation for my contribution to this study. I will receive the $ 25 CAD gift card, regardless of continuation in the study.

Voluntary Participation: I am under no obligation to participate and if I choose to participate, I can withdraw from the study at any time and/or refuse to answer any questions, without suffering any negative consequences. If I choose to withdraw, all data gathered until the time of withdrawal will be destroyed unless I give permission for it to be used.

Acceptance: I, -- agree to participate in the above research study conducted by Rosiane Simeon of the Interdisciplinary School of Health Sciences, Faculty of Health Sciences, University of Ottawa, which research is under the supervision of Dr. Vivian A. Welch and Dr. Peter Tugwell.

If I have any questions about the study, I may contact the researcher or her supervisors.

If I have any questions regarding the ethical conduct of this study, I may contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 154, Ottawa, ON K1N 6N5
Tel.: (613) 562-5387
Email: ethics@uottawa.ca

There are two copies of the consent form, one of which is mine to keep.

Participant's signature: -- Date: -----------------------

Researcher's signature:--- Date: ----------------------------

Consent form – HTA analysts

Title of the study: Patient engagement for equity-focused health technology assessment (HTA) recommendations in the digital era

Principal Investigator: Rosiane Simeon, MPH, PhD (c)
Interdisciplinary School of Health Sciences, Faculty of Health Sciences, University of Ottawa
Phone: email:

Supervisors: Dr. Vivian Welch: and Dr. Peter Tugwell

School of Epidemiology and Public Health, Faculty of Medicine, University of Ottawa

Invitation to Participate: I am invited to participate in the abovementioned research study conducted by Rosiane Simeon, a PhD candidate supervised by Dr. Vivian Welch and Dr. Peter Tugwell.

Purpose of the Study: The purpose of the study is to assess the use of digital technologies for patient engagement to promote the inclusion and participation of patients with diverse background for informing the development of HTA recommendations that are reasonable and reflect the preferences of patients.

Participation: My participation will consist essentially of doing an interview with the principal investigator- Rosiane Simeon. The interview will take approximately 45-60 minutes during which I will share my experiences and perspectives about the use of digital technologies for inclusive and diverse patient engagement to inform HTA recommendations. I will communicate with the principal investigator to find a suitable date and time for the interview, which will take place online via Zoom. The interview will be audio recorded only.

Risks: My participation in this study will entail that I share my name, my title and specific role(s), the name of my affiliated organization and information on my personal experiences about participating in patient engagement activities. The researcher does not anticipate any major risks associated with me sharing the above information. The information that I share about my experiences will not be associated with me and my name will not be included in the publication of the results; only the researcher will have access to this information.

Benefits: My participation in this study will help contribute to the advancement of knowledge for developing patient engagement strategies that are inclusive of patients from diverse background, and that consider patients' perspectives in HTA recommendations.

Confidentiality and anonymity: I have received assurance from the researcher that the information I will share will remain strictly confidential.
I understand that the content will be used only to identify factors for the use of digital technologies to support patient engagement strategies and that my confidentiality will be protected and that no one other than the researcher will have access to the recording.

Anonymity will be protected in the following manner: interviews will be audio recorded only each interview will be de-identified and labelled using a unique alpha-numeric. All interviews will be transcribed by the principal investigator.

Written quotations of comments will be used in publications but de-identified when possible.

I understand that it might be impossible to completely conceal the identity of HTA organizations because it is a small community of research, where most organizations share knowledge and practices. Some organizations may be identifiable based on quotation content.

I understand that it might also be difficult to completely conceal the identity of CEOs because it is a high visibility role; and that CEOs may provide information that only them could know. This could make it easier to identify CEOs compared to a practitioner or general members of the organization.

I therefore selected Yes or No in the following options:

5. [] Yes, the researcher can use my title(s) in quotes (e.g. CEO)
6. [] No, the researcher cannot use my title(s) in quotes (e.g. participant 23B)
7. [] Yes, the researcher can link me to my organization (e.g. CEO of X organization)
8. [] No, the researcher cannot link me to my organization (e.g. CEO of unnamed organization)

Conservation of data: The data collected recordings and transcripts of interviews will be kept in a secure manner using alpha-numeric passwords on the principal investigator’s computer. All data will be stored at the principal investigator’s home for 5 years and will be accessed only by the principal investigator.

Compensation: There is no compensation for participating in this study.

Voluntary Participation: I am under no obligation to participate and if I choose to participate, I can withdraw from the study at any time and/or refuse to answer any questions, without suffering any negative consequences. If I choose to withdraw, all data gathered until the time of withdrawal will be destroyed unless I give permission for it to be used.

Acceptance: I, -- agree to participate in the above research study conducted by Rosiane Simeon of the Interdisciplinary School of Health Sciences, Faculty of Health Sciences, University of Ottawa, which research is under the supervision of Dr. Vivian A. Welch and Dr. Peter Tugwell.

If I have any questions about the study, I may contact the researcher or her supervisors.

If I have any questions regarding the ethical conduct of this study, I may contact the Protocol Officer for Ethics in Research, University of Ottawa, Tabaret Hall, 550 Cumberland Street, Room 154, Ottawa, ON K1N 6N5

Tel.: (613) 562-5387
Email: ethics@uottawa.ca

There are two copies of the consent form, one of which is mine to keep.

Participant's signature: -- Date: -----------------------

Researcher's signature:-- Date: ----------------------------

Appendix D: Interview guides

INTERVIEW GUIDE – PATIENTS

Introduction

You have agreed to participate in a qualitative study to share your perspectives and your experiences in patient engagement activities to inform HTA recommendations.

As indicated in the consent form, this study aims **to identify implementation considerations for the use of digital technologies in HTA-related patient engagement activities for improving participation and inclusion of diverse population groups**. I will be recording the interviews with the **Audio Record** feature of the Zoom application. I will be taking some notes as well. Note that I will not use the Video feature.

Interviews will be transcribed and de-identified - no one you mention will be identifiable by their names in the transcripts. Then I will repeatedly analyze the text to look for both common themes and perspectives between respondents as well as examples of disagreement.

I intend to disseminate the findings as abstracts in HTA-related conferences and publish the findings in the peer-reviewed literature. Written quotations of comments will be used but de-identified.

The interview will take approximately 45-60 minutes. Before we begin, I would like to remind you that there are no correct answers. Your participation is **voluntary**. You do not have to answer any questions that make you feel uncomfortable. You can choose to end the interview at any point.

Do you have any questions about the study and the use of data before I begin recording?

Questions For Patient representatives

Part 1: Brief description of your patient organization(s):

1. Could you please briefly describe your organization?

Probes:

- Could you please tell me about the objectives of your organizations?
- Could you tell me what is the biggest challenge that your organization face in terms of:
    - Recruiting members with diverse background or needs,
    - Reaching out to its members
    - Ensuring meaningful participation in patient engagement activities within the organization
    - Bringing their voices forward
- Could you please tell me more about the structure of your organization: roughly, how many members are involved in your organization?
- Which health conditions are concerned?
- What is your geographical coverage?
    - Is it a local organization/national/international organization?

Part 2: Brief description of your roles and responsibilities

Could you please tell me about your involvement in patient engagement for HTA?

Probes

- How long have you been involved in patient engagement activities for HTA?
    - Which institutions have you been working with?
- Could you please tell me about your different roles and responsibilities related to your involvement in patient engagement activities?
    - Have these roles changed over time?
    - What have been your roles/responsibilities within your own patient organization?
    - What have been your roles/responsibilities in collaborating with HTA organizations on behalf of your organization?
        - Could you please elaborate more on your experience with that

Part 3: Description of the use of digital technologies for patient engagement activities

3. Could you please share your experiences with the use of digital technologies (i.e. phone, email, social media- Facebook, Twitter, Skype, website, etc.) to communicate and collect information for patient engagement activities within your own organization?

Probes

- How do you use digital technologies to gather feedback / comments/ perspectives from your organization's members on health technologies or their health conditions?
  - What have been your own experience to in using these – webinars, blogs, photo voice, social media pages, discussion group, forums, surveys etc. to communicate with members?
- How do you use digital technologies when collaborating with HTA organizations?
  - What have been your own experience in using these media – webinars, blogs, photo voice, social media pages, discussion group, forums, surveys etc. to provide input to HTA organizations?
  - What types of digital technologies do you generally use to provide input to HTA organization? (emails, telephones, blogs, webinar software, social media-Facebook, Twitter, Skype, etc.,)
- Can you elaborate more on your experience with using these digital technologies to provide input to HTA organizations?
  - How did you provide input to HTA organizations?
    - Have you participated in advisory panel, responded to survey, commented on documents?

Part 4: Identification of implementation considerations for the use of digital technologies to increase participation and inclusion of diverse groups in patient engagement activities

4. a: In what ways do you think your organization could use digital technologies to achieve its objectives related to patient engagement?

Probes

- What are some ways you think your organization could use digital technologies to increase participation and encourage meaningful engagement amongst its members?

4. b: How do you think your organization could use digital technologies to improve the inclusion of members with diverse background, needs, living in different locations?

Probes

- Could you please tell me about how your organization could use digital technologies to identify, recruit new members or reach out to existing members?

Part 5: General questions about challenges to using digital technologies for patient engagement in HTA and potential solutions to those challenges

5a. Could you please tell me what you consider to be the biggest challenges to using digital technologies for patient engagement? Within your own organization? or between patient and HTA organizations?

Probes

- Could you please provide some examples of barriers that you think are particularly significant when implementing a digital strategy for engagement within your organization?
  - Why is this challenge so significant?
  - Are there any other challenges or barriers that you think are particularly important to consider?

5.b. Could you think of some ways your organization could use to overcome these barriers?

Probes

a. Can you give me some examples of solutions to address these challenges?
  - What exactly you think needs to be done?

- What would you say make a digital strategy for patient engagement a great success for your organization?

5.c. Could you tell me how your organization and its members could work to make a digital strategy for patient engagement sustainable?

Part 6: Additional comments/identification of other potential research participants

6. Would you like to add anything?

Probes

- Anything that we haven’t talked about that you think important and potentially relevant to the use of digital technologies for inclusive patient engagement?

Closing comments

Thank you for your time. Do you have any questions for me?

INTERVIEW GUIDE – HTA ORGANIZATIONS

Introduction

You have agreed to participate in a qualitative study to share your perspectives and your experiences in patient engagement activities to inform HTA recommendations.

As indicated in the consent form, this study aims **to identify implementation considerations for the use of digital technologies in HTA-related patient engagement activities for improving participation and inclusion of diverse population groups**. I will be recording the interviews with the **Audio Record** feature of the Zoom application. I will be taking some notes as well. Note that I will not use the Video feature.

Interviews will be transcribed and de-identified - no one you mention will be identifiable by their names in the transcripts. Then I will repeatedly analyze the text to look for both common themes and perspectives between respondents as well as examples of disagreement.

I intend to disseminate the findings as abstracts in HTA-related conferences and publish the findings in the peer-reviewed literature. Written quotations of comments will be used but de-identified.

The interview will take approximately 45-60 minutes. Before we begin, I would like to remind you that there are no correct answers. Your participation is **voluntary**. You do not have to answer any questions that make you feel uncomfortable. You can choose to end the interview at any point.

Do you have any questions about the study and the use of data before I begin recording?

Questions

For HTA Practitioners/CEO of HTA organizations

Part 1: Brief description of your HTA organization(s):

2. Could you please briefly describe your organization?

Probes:

- Could you please tell me about your organization's patient engagement strategy?
- Could you tell me what is the biggest challenge that your organization face in terms of:
  - Ensuring meaningful participation in patient engagement activities
- Could you please tell me more about the structure of your organization's jurisdiction?
  - What is your geographical coverage?
  - Is it a local organization/national/international organization?

Part 2: Brief description of your roles and responsibilities

2.a. Could you please tell me about your involvement in HTA?

Probes

- Could you please tell me about your different roles and responsibilities related to your involvement in HTA?
  - Have these roles changed over time?

2.b. Have you been involved in patient engagement activities to inform your work?

Probes

- How long have you been involved in patient engagement activities for HTA?
  - Which institutions have you been working with?
  - What have been your roles/responsibilities in collaborating with HTA patient organizations on behalf of your organization?
    - Could you please elaborate more on your experience with that?

Part 3: Description of the use of digital technologies for patient engagement activities

4. Could you please share your experiences with the use of digital technologies (i.e. phone, email, social media- Facebook, Twitter, Skype, website, etc.) to communicate with patient representatives and collect patient input?

- How do you use digital technologies to gather feedback / comments/ perspectives from patients on health technologies or their health conditions?
    - What have been your own experience to sing these media – webinars, blogs, photo voice, social media pages, discussion group, forums, surveys etc. to communicate with patients?
- How do you use digital technologies when collaborating with patient organizations?
    - What have been your own experience in using these media – webinars, blogs, photo voice, social media pages, discussion group, forums, surveys etc. to provide input to HTA organizations?
    - What types of digital technologies do you generally use to collect patient input? (emails, telephones, blogs, webinar software, social media-Facebook, Twitter, Skype, etc.,)
- Can you elaborate more on your experience with using these digital technologies to provide input to HTA organizations?
    - How did you provide input to HTA organizations?
        - Have you participated in advisory panel, responded to survey, commented on documents?

Part 4: Identification of implementation considerations for the use of digital technologies to increase participation and inclusion of diverse groups in patient engagement activities

4. a: In what ways do you think your organization could use digital technologies to achieve its objectives related to patient engagement?

Probes

- What are some ways you think your organization could use digital technologies to increase participation and promote meaningful patient engagement?

4. b: How do you think your organization could use digital technologies to improve the inclusion of patients with diverse background, needs, living in different locations to inform your work?

Probes

- Could you please tell me about how your organization could use digital technologies to identify, recruit or reach out to patients?

Part 5: General questions about challenges to using digital technologies for patient engagement in HTA and potential solutions to those challenges

5a. Could you please tell me what you consider to be the biggest challenges to using digital technologies for patient engagement?

Probes

- Could you please provide some examples of barriers that you think are particularly significant when implementing a digital strategy for engagement within your organization?
  - Why is this challenge so significant?
  - Are there any other challenges or barriers that you think are particularly important to consider?

5.b. Could you think of some ways your organization could use to overcome these barriers?

Probes

b. Can you give me some example of solutions to address these challenges?
   - What exactly you think needs to be done?

- What would you say make a digital strategy for patient engagement a great success for your organization?

5.c. Could you tell me how your organization and its members could work to make a digital strategy for patient engagement sustainable?

Part 6: Additional comments/identification of other potential research participants

6. Would you like to add anything?

Probes

- Anything that we haven't talked about that you think important and potentially relevant to the use of digital technologies for inclusive patient engagement?

Closing comments

Thank you for your time. Do you have any questions for me?

Appendix E: Additional illustrative quotes- Patients

Themes	Sub-themes
Perceptions about the Role of patient engagement in the development of Equity-focused HTA recommendations	
• Importance of patient engagement to the HTA process • Patient engagement as the unique source of evidence • Meaning /implications of health equity • Limitations of current health equity analysis in HTA (Challenges)	*“We use patient engagement as a kind of process to advocate for programs, policies and directives that cover the real needs of patients. We worked hard to make sure that patient concerns are taken seriously.” [Patient 04]* *“That was very much the message I brought to them: you cannot ignore this[autism] population now and over time.” [Patient 05]*
Perceptions about diverse and inclusive patient engagement strategies in HTA	
• Expectations about diverse and inclusive patient engagement strategies • Positive experiences with current engagement processes/ inclusive engagement	*Ensuring we're [patient organizations] hearing from equity-seeking groups is challenging. And one of the issues is that there's not maybe enough diversity in the people reaching out to them. And so, the faces of the researchers must change. People realize that more as we talk about [health] equity.” [Patient 01*
• Build trust in the HTA process • Cultural competencies • Provide practical support to patients	*So, until the system understands the importance of cultural competencies, there will be no equity in healthcare delivery [patient 04]* *“HTA organizations need some form of reimbursement [for patient engagement] because we're already at a detriment, not being able to have enough accessible jobs... there's so few jobs for people with disabilities.” [Patient 02]* The following interviewee shared how their try to bring the perspectives of other patients in patient engagement: *“I have been diagnosed with several autoimmune diseases, and I looked for ways to engage in the health system. And in part, because I knew other*

	patients like me could use the feedback in the system." [Patient 02]
Considerations for implementing digitally enabled patient engagement strategies for equity-focused HTA recommendations.	
• Characteristics of digital technologies for inclusion of people with different abilities • Advantages of digitally-enabled engagement (Perceived usefulness) • Advantages of in-person meetings • Practical and ethical issues in using digital technologies (Digital divide) • Mixed modalities for diverse and inclusive engagement in HTA (Recommendations)	*The shift in using digital technologies has clear benefits. I don't have to leave my house because this causes stress and anxiety. So, this way, I can participate." [Patient 03]* *"Digital technology would be interesting to bring patients even closer to advisory committee meetings… The fear of bringing people in person to speak to a committee is that people are fighting for life-saving drugs." [Patient 01]*

Appendix F: Illustrative quotes- HTA analysts

Patient engagement as critical source of evidence in HTA	
Subthemes	*Quotes*
Identify health equity concerns	*"Patient engagement help understand those experiences of patients and families, their burdens, their costs that they're taking on their position when it comes to accessing treatment, and maybe what barriers are there to accessing a treatment." HTA analyst 06* *"Patients are very keen to share their experience because they felt like that no one really understood the reality that they face and not just the reality in terms of like a health outcome, what it was like to go through that journey." HTA analyst 01*
Drivers of implementing patient engagement to inform health equity analysis in HTA	
Patient engagement is not always necessary for health equity analysis (learning curve)	*"We've done other ones where the technology was software on the computer; we didn't do patient engagement because the patient has no input or knowledge about it anyway." HTA analyst 03* *"I can almost see an equity angle to almost any type of health technology. But whether patient engagement is the way to understand that equity consideration. that is not always the case." HTA analyst 06* *"The technologies were so niche. [we] didn't have a big selection pool of people talk to." HTA analyst 01*
Importance of health equity in HTA (learning curve)	*"Until recently health equity was probably not a major focus of our HTAs. It was included on the periphery". HTA analyst 03* *"it would be broadly challenging to say that there is a health technology that doesn't require equity considerations, because you are privileging certain people in the very decision that you're being asked to make." HTA analyst 02* *"When you talk to patients, and then they'll tell you in as much as you're interested in measuring my blood sugar or whatever, what's important for me is my quality of life". HTA analyst 05*
Data availability (Statistical representation	*"It can be hard to identify equity concerns if the original authors didn't do it[health equity analysis], and this will impact patient*

	engagement because we don't know whose perspective should we be seeking and who should we be talking to...” HTA analyst 02 *Solution: “If Health Canada and the FDA as regulatory bodies then said you pharmaceutical companies have to put a component of equity into[their research, before we actually approve it..,?” HTA analyst 04*
Data availability (Symbolic representation	*Solution: “Even outside of the minority groups, sometimes it is hard to find patient representatives for certain conditions, I’m hoping we will get to a stage where whoever is representing is not representing for themselves, they're representing for everybody.” HTA analyst 05*
Learning curve	*“It is also the language we use… We're learning how to reflect it in the recommendation and so it's a learning curve.” HTA analyst 04*
HTA timeline (capacity building)	*“One challenge is the timeline. We need to get the assessments done quickly. So, it doesn't leave a lot of time for the kind of reflection that you would need to do on what you're reading in the patient input” HTA analyst 06*
Application of framework (Characterization and reporting)	*“I mean, in truth I was struggling with how to position this. And then I said, let's go back and read the equity framework, because this is an equity thing right here this people who can't use [this technology] are not getting the same outcomes as people who can…if they have access to [this other technology] then they would be afforded the same outcomes as their peers… you can see the learning that we applied, you can see equity thinking, right? HTA analyst 04* *“Reporting using a framework [means that] there is a box for equity, you have to fill it in, if you don't fill it in, and then you haven't done justice to the process and you're not using the framework appropriately” HTA analyst 05.* *Limitation: “Some aspects of equity in the PROGRESS checklist are more relevant than others. There's some disadvantage that's caused by the technology… So that's another example of how we look for*

	equity in their story, rather than always explicitly asking about it." HTA analyst 06
Training (capacity building)	*"Training for the staff so that they know what that [health equity analysis] might mean for different groups, disadvantaged groups, or equity considerations; it really depends on the capabilities of the reviewer, whether it [health equity] makes it into the report or not." HTA analyst 06*
Implication of equity-focused HTA recommendations (health equity analysis)	*""To implement an [equity-focused] recommendation requires collaboration among different ministry areas. Systems work better in silos from an organized perspective... systems don't work good in silos from an equitable perspective" HTA analyst 04*
5.1.Mandates of patient organizations and health equity (future research)	*"When it comes to equity, then we might have to ask [patient groups]..even if they're small groups or big groups, but if their mandate is access to the product, they may be less likely to emphasize the equity aspect. Because they want to show that it's beneficial to have it, you know, they have a certain position, whereas other groups don't have a position when it comes to drug access." HTA analyst 06*
Conflict of interest (not reporting in analysis-future research)	*"This is personal and not organizational… I certainly understand why [patient organizations] would use industry funding to support the work that they're doing…there shouldn't be any sort of judgment around the funding source of the patient groups that are providing inputs, as long as they are open [about it]" HTA analyst 02*

Additional files

Final R stats 2023-04-04

```
library(readxl)
```

```
## Warning: package 'readxl' was built under R version 4.2.3
```

```
HTA2022_Final_data <- read_excel("HTA2022_Final_Bin2_ 4April2023.xlsx")
attach(HTA2022_Final_data)
```

Model 1 - Add few words here

```
## Model EquityRec  ~ PatEngInd
EqPatEngInd <- glm(EquityRec  ~ PatEngInd, family = "gaussian")
# model summary
summary(EqPatEngInd)
```

```
## 
## Call:
## glm(formula = EquityRec ~ PatEngInd, family = "gaussian")
## 
## Deviance Residuals: 
##    Min      1Q  Median      3Q     Max  
##  -1.10   -0.75   -0.10    0.25    2.25  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   2.1000     0.1932  10.869 1.29e-15 ***
## PatEngInd    -1.3500     0.2366  -5.705 4.15e-07 ***
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 0.7465517)
## 
##     Null deviance: 67.6  on 59  degrees of freedom
## Residual deviance: 43.3  on 58  degrees of freedom
## AIC: 156.7
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqPatEngInd)
```

```
##                 2.5 %     97.5 %
## (Intercept)  1.721328  2.4786719
## PatEngInd   -1.813776 -0.8862236
```

```
# Odds ratio
exp(EqPatEngInd$coefficients[-1])
```

```
## PatEngInd
## 0.2592403
```

```
# Confidence interval for odds ration
exp(confint.default(EqPatEngInd))
```

```
##                  2.5 %     97.5 %
## (Intercept) 5.5919504 11.9254154
## PatEngInd   0.1630373  0.4122095
```

Model 2 -

```
## Model EquityRec  ~ HTARev
Review <- glm(EquityRec  ~ HTARev, family = "gaussian")
# model summary
summary(EqReview)
```

```
##
## Call:
## glm(formula = EquityRec ~ HTARev, family = "gaussian")
##
## Deviance Residuals:
##     Min       1Q   Median       3Q      Max
## -1.1422  -0.5452  -0.1422   0.1639   2.5518
##
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)
## (Intercept)   0.4482     0.2108   2.126   0.0378 *
## HTARev        0.6940     0.1594   4.355  5.5e-05 ***
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
##
## (Dispersion parameter for gaussian family taken to be 0.8783548)
##
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 50.945  on 58  degrees of freedom
## AIC: 166.46
##
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqReview)
```

```
##                  2.5 %    97.5 %
## (Intercept) 0.03498481 0.8614007
## HTARev      0.38162005 1.0063318
```

```
# Odds ratio
exp(EqReview$coefficients[-1])
```

```
##   HTARev
## 2.001658
```

```
# Confidence interval for odds ration
exp(confint.default(EqReview))
```

```
##                2.5 %   97.5 %
## (Intercept) 1.035604 2.366473
## HTARev      1.464655 2.735548
```

Model 3 -

```
## Model EquityRec  ~ DecMak
EqDecM <- glm(EquityRec  ~ DecMak, family = "gaussian")
# model summary
summary(EqDecM)
```

```
##
## Call:
## glm(formula = EquityRec ~ DecMak, family = "gaussian")
##
## Deviance Residuals:
##     Min       1Q   Median       3Q      Max
## -1.5429  -0.7200   0.2800   0.4571   2.4571
##
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)
## (Intercept)   1.5429     0.1686   9.149 7.53e-13 ***
## DecMak       -0.8229     0.2612  -3.150  0.00258 **
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
##
## (Dispersion parameter for gaussian family taken to be 0.9952709)
##
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 57.726  on 58  degrees of freedom
## AIC: 173.95
##
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqDecM)
```

```
##                 2.5 %     97.5 %
## (Intercept)  1.212347  1.8733672
## DecMak      -1.334881 -0.3108331
```

```
# Odds ratio
exp(EqDecM$coefficients[-1])
```

```
##    DecMak
## 0.4391751
```

```
# Confidence interval for odds ration
exp(confint.default(EqDecM))
```

```
##                 2.5 %    97.5 %
## (Intercept) 3.3613647 6.5101809
## DecMak      0.2631894 0.7328362
```

Model 4 -

```
## Model EquityRec  ~ HTAYr
EqYr <- glm(EquityRec  ~ HTAYr, family = "gaussian")
# model summary
summary(EqYr)
```

```
##
## Call:
## glm(formula = EquityRec ~ HTAYr, family = "gaussian")
##
## Deviance Residuals:
##     Min       1Q   Median       3Q      Max
## -1.2941  -0.6667  -0.2941   0.7059   2.7059
##
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)
## (Intercept)   0.6667     0.3518   1.895    0.063 .
## HTAYr         0.6275     0.3815   1.645    0.105
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
##
## (Dispersion parameter for gaussian family taken to be 1.11359)
##
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 64.588  on 58  degrees of freedom
## AIC: 180.69
##
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqYr)
```

```
##                   2.5 %   97.5 %
## (Intercept) -0.02276234 1.356096
## HTAYr       -0.12033976 1.375242
```

```
# Odds ratio
exp(EqYr$coefficients[-1])
```

```
##    HTAYr
## 1.872831
```

```
# Confidence interval for odds ration
exp(confint.default(EqYr))
```

```
##                   2.5 %   97.5 %
## (Intercept) 0.9774948 3.881011
## HTAYr       0.8866191 3.956033
```

Model 5 -

```
## Model EquityRec  ~ HTAYrEI
EqYrEI <- glm(EquityRec  ~ HTAYrEI, family = "gaussian")
# model summary
summary(EqYrEI)
```

```
## 
## Call:
## glm(formula = EquityRec ~ HTAYrEI, family = "gaussian")
## 
## Deviance Residuals: 
##     Min       1Q   Median       3Q      Max  
## -1.2941  -0.6667  -0.2941   0.7059   2.7059  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   1.2941     0.1478   8.758 3.34e-12 ***
## HTAYrEI      -0.6275     0.3815  -1.645    0.105    
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 1.11359)
## 
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 64.588  on 58  degrees of freedom
## AIC: 180.69
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqYrEI)
```

```
##                 2.5 %    97.5 %
## (Intercept)  1.004500 1.5837358
## HTAYrEI     -1.375242 0.1203398
```

```
# Odds ratio
exp(EqYrEI$coefficients[-1])
```

```
##   HTAYrEI 
## 0.5339511
```

```
# Confidence interval for odds ration
exp(confint.default(EqYrEI))
```

```
##                  2.5 %   97.5 %
## (Intercept) 2.7305404 4.873127
## HTAYrEI     0.2527785 1.127880
```

Model 6 -

```
## Model EquityRec  ~ pCODR
EqpCODR <- glm(EquityRec  ~ pCODR, family = "gaussian")
# model summary
summary(EqpCODR)
```

```
## 
## Call:
## glm(formula = EquityRec ~ pCODR, family = "gaussian")
## 
## Deviance Residuals: 
##     Min       1Q   Median       3Q      Max  
## -1.3333  -0.8000  -0.3333   0.6667   2.6667  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   1.3333     0.1571   8.488 9.36e-12 ***
## pCODR        -0.5333     0.3142  -1.698   0.0949 .  
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 1.110345)
## 
##     Null deviance: 67.6  on 59  degrees of freedom
## Residual deviance: 64.4  on 58  degrees of freedom
## AIC: 180.52
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqpCODR)
```

```
##                  2.5 %     97.5 %
## (Intercept)  1.025461 1.64120574
## pCODR       -1.149078 0.08241149
```

```
# Odds ratio
exp(EqpCODR$coefficients[-1])
```

```
##     pCODR 
## 0.5866462
```

```
# Confidence interval for odds ration
exp(confint.default(EqpCODR))
```

```
##                 2.5 %   97.5 %
## (Intercept) 2.7883804 5.161389
## pCODR       0.3169288 1.085903
```

Model 7 -

```
## Model EquityRec  ~ CDR
EqCDR <- glm(EquityRec  ~ CDR, family = "gaussian")
# model summary
summary(EqCDR)
```

```
## 
## Call:
## glm(formula = EquityRec ~ CDR, family = "gaussian")
## 
## Deviance Residuals: 
##     Min       1Q   Median       3Q      Max  
## -1.5429  -0.7200   0.2800   0.4571   2.4571  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   1.5429     0.1686   9.149 7.53e-13 ***
## CDR          -0.8229     0.2612  -3.150  0.00258 ** 
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 0.9952709)
## 
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 57.726  on 58  degrees of freedom
## AIC: 173.95
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqCDR)
```

```
##                  2.5 %     97.5 %
## (Intercept)  1.212347  1.8733672
## CDR         -1.334881 -0.3108331
```

```
# Odds ratio
exp(EqCDR$coefficients[-1])
```

```
##       CDR 
## 0.4391751 
```

```
# Confidence interval for odds ration
exp(confint.default(EqCDR))
```

```
##                 2.5 %    97.5 %
## (Intercept) 3.3613647 6.5101809
## CDR         0.2631894 0.7328362
```

Model 8 -

```
## Model EquityRec  ~ HQO
EqHQO <- glm(EquityRec  ~ HQO, family = "gaussian")
# model summary
summary(EqHQO)
```

```
## 
## Call:
## glm(formula = EquityRec ~ HQO, family = "gaussian")
## 
## Deviance Residuals: 
##    Min      1Q  Median      3Q     Max  
##  -1.10   -0.75   -0.10    0.25    2.25  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   0.7500     0.1366   5.490 9.29e-07 ***
## HQO           1.3500     0.2366   5.705 4.15e-07 ***
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 0.7465517)
## 
##     Null deviance: 67.6  on 59  degrees of freedom
## Residual deviance: 43.3  on 58  degrees of freedom
## AIC: 156.7
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqHQO)
```

```
##                 2.5 %   97.5 %
## (Intercept) 0.4822386 1.017761
## HQO         0.8862236 1.813776
```

```
# Odds ratio
exp(EqHQO$coefficients[-1])
```

```
##      HQO 
## 3.857426
```

```
# Confidence interval for odds ration
exp(confint.default(EqHQO))
```

```
##                 2.5 %   97.5 %
## (Intercept) 1.619696 2.766994
## HQO         2.425951 6.133567
```

Model 9 -

```
## Model EquityRec  ~ PatEngDir
EqPatEngDir <- glm(EquityRec  ~ PatEngDir, family = "gaussian")
# model summary
summary(EqPatEngDir)
```

```
##
## Call:
## glm(formula = EquityRec ~ PatEngDir, family = "gaussian")
##
## Deviance Residuals:
##    Min      1Q  Median      3Q     Max
##  -1.10   -0.75   -0.10    0.25    2.25
##
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)
## (Intercept)   0.7500     0.1366   5.490 9.29e-07 ***
## PatEngDir     1.3500     0.2366   5.705 4.15e-07 ***
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
##
## (Dispersion parameter for gaussian family taken to be 0.7465517)
##
##     Null deviance: 67.6  on 59  degrees of freedom
## Residual deviance: 43.3  on 58  degrees of freedom
## AIC: 156.7
##
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqPatEngDir)
```

```
##                  2.5 %   97.5 %
## (Intercept) 0.4822386 1.017761
## PatEngDir   0.8862236 1.813776
```

```
# Odds ratio
exp(EqPatEngDir$coefficients[-1])
```

```
## PatEngDir
##  3.857426
```

```
# Confidence interval for odds ration
exp(confint.default(EqPatEngDir))
```

```
##                 2.5 %   97.5 %
## (Intercept) 1.619696 2.766994
## PatEngDir   2.425951 6.133567
```

Model 10 (on the pdf file) is Model 1

Model 11 -

```
## Model EquityRec  ~ DecMakC
EqDecMakC <- glm(EquityRec  ~ DecMakC, family = "gaussian")
# model summary
summary(EqDecMakC)
```

```
## 
## Call:
## glm(formula = EquityRec ~ DecMakC, family = "gaussian")
## 
## Deviance Residuals: 
##     Min       1Q   Median       3Q      Max  
## -1.5429  -0.7200   0.2800   0.4571   2.4571  
## 
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)    
## (Intercept)   0.7200     0.1995   3.609 0.000643 ***
## DecMakC       0.8229     0.2612   3.150 0.002583 ** 
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
## 
## (Dispersion parameter for gaussian family taken to be 0.9952709)
## 
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 57.726  on 58  degrees of freedom
## AIC: 173.95
## 
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqDecMakC)
```

```
##                  2.5 %   97.5 %
## (Intercept) 0.3289352 1.111065
## DecMakC     0.3108331 1.334881
```

```
# Odds ratio
exp(EqDecMakC$coefficients[-1])
```

```
##  DecMakC 
## 2.276996 
```

```
# Confidence interval for odds ration
exp(confint.default(EqDecMakC))
```

```
##                 2.5 %   97.5 %
## (Intercept) 1.389488 3.037591
## DecMakC     1.364561 3.799544
```

Model 12 -

```
## Model EquityRec  ~ DecMakV
EqDecMakV <- glm(EquityRec  ~ DecMakV, family = "gaussian")
# model summary
summary(EqDecMakV)
```

```
##
## Call:
## glm(formula = EquityRec ~ DecMakV, family = "gaussian")
##
## Deviance Residuals:
##     Min       1Q   Median       3Q      Max
## -1.5429  -0.7200   0.2800   0.4571   2.4571
##
## Coefficients:
##             Estimate Std. Error t value Pr(>|t|)
## (Intercept)   1.5429     0.1686   9.149 7.53e-13 ***
## DecMakV      -0.8229     0.2612  -3.150  0.00258 **
## ---
## Signif. codes:  0 '***' 0.001 '**' 0.01 '*' 0.05 '.' 0.1 ' ' 1
##
## (Dispersion parameter for gaussian family taken to be 0.9952709)
##
##     Null deviance: 67.600  on 59  degrees of freedom
## Residual deviance: 57.726  on 58  degrees of freedom
## AIC: 173.95
##
## Number of Fisher Scoring iterations: 2
```

```
# Confidence interval of coefficients
confint.default(EqDecMakV)
```

```
##                 2.5 %     97.5 %
## (Intercept)  1.212347  1.8733672
## DecMakV     -1.334881 -0.3108331
```

```
# Odds ratio
exp(EqDecMakV$coefficients[-1])
```

```
##   DecMakV
## 0.4391751
```

```
# Confidence interval for odds ration
exp(confint.default(EqDecMakV))
```

```
##                 2.5 %    97.5 %
## (Intercept) 3.3613647 6.5101809
## DecMakV     0.2631894 0.7328362
```