A STUDY OF RESEARCH PRIORITY SETTING FOR MYELODYSPLASTIC SYNDROMES IN CANADA

Dr. Grace Christou, MD, MSc (Biochemistry), FRCP(C)

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

Preface

The following is a manuscript-based thesis that has been performed as part of a Master of Science in Clinical Epidemiology degree at the University of Ottawa. It has been formatted in accordance with guidance set out by the intended peer-reviewed journals.

Ethics approval was needed and obtained for the second manuscript from the OHSN REB. The first article, 'Sixteen Years of James Lind Alliance Priority Setting Partnerships: A Scoping Review of Implementation Strengths and Challenges' has not been submitted for publication yet but is formatted for *BMJ Open*. I designed and carried out the study in its entirety. Collaboration from co-authors was provided for literature search (Risa Shorr) and coding verification (Dr. Ian Graham). The second paper, 'Research Priorities in Myelodysplastic Syndromes: A Survey of Patients, Caregivers and Clinicians' has been formatted but not yet submitted to *American Journal of Hematology.* I designed this study in its entirety with feedback and guidance from supervisors and co-authors. Dr. Deepro Chowdhury contributed to coding verification.

I certify that I am and will be the first and corresponding author on each of these papers. I wrote the papers with guidance from supervisors and co-authors and will be the primary author to review and address feedback from peer reviewers.

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List of Abbreviations

AAMAC: Aplastic Anemia and Myelodysplasia Association of Canada

ADHD: Attention Deficit and Hyperactivity Disorder

CHERRIES: The Checklist for Reporting Results of Internet E-Surveys

DM: Diabetes Mellitus

EB: Epidermolysis Bullosa

FREE TEA: Facilitating Representative and EquitablE involvement through Targeted Engagement and Assisted involvement

HCP: Health care provider

JBI: Joanna Briggs Institute

JLA: James Lind Alliance

LGBTQ2: Lesbian, Gay, Bisexual, Transgender, Queer and 2 Spirited

MDS: Myelodysplastic Syndromes

PEI: Prince Edward Island

PICO: Population, Intervention, Comparator, Outcome

PRESS: Peer Review of Electronic Search strategy

PRISMA-ScR: Preferred Reporting Items for SysteMAtic Reviews – Scoping Reviews

PSP: Priority Setting Partnership

Thesis Abstract

The James Lind Alliance approach is a widely used method for research priority setting, yet comprehensive reviews of its implementation are lacking. This thesis therefore focused on reviewing and analyzing the barriers and facilitators to its implementation and on carrying out the early stages of our own priority setting partnership to prioritize research in myelodysplastic syndromes (MDS).

A scoping review of the barriers and facilitators encountered when undertaking a James Lind Alliance priority setting partnership was conducted. Our analysis identified that most barriers could be categorized into three main areas: representation, data collection and processing and result useability. We also identified a number of methods that were used successfully to overcome some of the barriers and offered our suggestions for further study.

Along with the Aplastic Anemia and Myelodysplasia Association of Canada, we conducted the initial stages of the Canadian MDS Priority Setting Partnership. We were able to collect 206 relevant research questions from people living with MDS, their caregivers and front line health professionals which can be put forward for further prioritization.

Chapter 1

Introduction

Thesis Purpose:

The purpose of my thesis is to systematically review the James Lind Alliance Priority Setting Partnerships' (PSP) process and to describe the conduct and results of the first stages of the Canadian Myelodysplastic Syndromes (MDS) PSP.

Misaligned Healthcare Research:

The problem of research waste, where research dollars are spent on poorly conceived, designed and conducted studies, has been recognized as a major barrier to improving human health and solving problems in healthcare. One of the areas that contribute to research waste is the misalignment between research activity and the priorities of end users, namely people suffering from medical conditions and the health care professionals directly caring for them [1]. In one of the clearest examples, a study examining the research priorities of patients with osteoarthritis found that only 9% of patients wanted more research into drugs, while the majority of patients and clinicians ranked more rigorous research of physiotherapy, surgery, education and coping strategies as their top priorities [2]. Despite that, 80% of clinical trials in osteoarthritis of the knee were drug evaluations[2]. Even within drug trials, the outcomes chosen by researchers are not always the most relevant to patients. An initiative to involve patients in the design of studies in rheumatoid arthritis for example, showed that the outcome most often used in trials to assess efficacy was pain, whereas fatigue was the most important symptom identified by patients [3]. A broader analysis of patient and clinician priorities across 14 health conditions demonstrated that while drugs constituted just 19% of patient and clinician research priorities, they remain the focus of 37% of non-commercial and 89% of industry trials [4].

This mismatch between the priorities of researchers and end-users has contributed to the perception that research does not serve to improve people's health, in turn leading to a diminished trust in, and uptake of, research findings[5,6].

A lack of patient involvement in research deprives researchers of an important source of lived expertise in their field of study. It is increasingly recognized that patients have a unique perspective of their condition which encompasses the experience of living with the condition and the social and economic context surrounding it, factors which play a critical role in the design and implementation of appropriate solutions. Thus, the inclusion of patient and clinicians' voices can enrich research by highlighting important knowledge gaps, identifying more impactful study outcomes and bringing forth recommendations that could improve study accrual and real-world implementation of findings[7].

In order to address this imbalance, public and private funders of research in many countries have developed frameworks for involving patients and point-of-care health care professionals in all stages of research. These frameworks provide clear guidelines on how to meaningfully include patients in research, and fostering mutual respect and trust between researchers and the public[8]. The cumulative effect is the conduct of more transparent, accountable, relevant, and impactful health research. Patient involvement in research priority setting is just one way in which research can become more inclusive, and within research priority setting, the James Lind Alliance is a commonly used framework and the focus of the thesis[8].

Myelodysplastic Syndromes:

Myelodysplastic syndromes (MDS) are a group of heterogeneous malignant clonal disorders of the bone marrow characterized by morphologic and functional abnormalities in blood cell production and low blood counts [9], leading to reduced functional capacity, recurrent serious infections and bleeding. It was reclassified in 2001 from a condition with low malignant potential to a cancer[10]. The presentation of MDS ranges from isolated mild cytopenias which may be stable for years to severe and rapidly fatal disease. In

all cases, a diagnosis of MDS carries a high risk for progression to acute myeloid leukemia (AML), a diagnosis that itself carries a high mortality and morbidity rate.

MDS has median age of diagnosis of 70 years and an incidence that rises sharply with age. In the general population, MDS has an incidence of 5 cases/100,000 persons/year, although the true incidence is estimated to be between 5 and 13 cases/100,000 annually[11] and the incidence among those over 60 is 5 to 6 times that of the general population [9,11].

Researcher Positionality:

As a clinical hematologist specializing in acute leukemia and myelodysplastic syndromes (MDS) and an early career researcher, MDS always seemed the most difficult of conditions to explain and communicate, and treatment options very individualized and highly dependent on patients' social context, personal values and functional status. I entered clinical fellowship training at a time when hypomethylating agents were becoming more widely used and our understanding of the molecular biology of MDS was growing rapidly. These changes have improved survival of patients with MDS. With that, came new questions from patients and caregivers about how new scientific insights impacted their future treatment options and how accepting active treatment will affect their quality of life. Patients on treatment asked how to mitigate persistent debilitating fatigue which persisted despite excellent blood counts. Many wondered how they could participate in maximizing quality of life and whether diet and exercise might enhance the effectiveness of pharmacologic treatment. Unfortunately, in most cases, these very important patient-centred questions are not addressed in the scientific literature. These questions about aspects of care beyond overall survival and progression free survival, helped me realize that people living with health conditions had important insights that could be instrumental to researchers' work in understanding disease biology and better evaluating novel treatments. It also showed me first hand that people living with terminal illness were heavily invested in getting the most out of their lives, something that perhaps should have been self-evident, but had not been at the forefront of my thinking until that point.

Coming face to face daily with these important evidence gaps, I decided I wanted to more robustly and systematically identify research gaps for this group of patients which lead me to the James Lynd Alliance method for research priority setting. As I explored how to use this method with patient with MDS and clinicians in Canada I was curious to know about the use of the JLA approach including the challenges encountered during its implementation and potential solutions.

The Need for Research Priority Setting In MDS:

MDS is curable only by allogeneic stem cell transplantation[12], where bone marrow from a healthy donor is transplanted in to a patient with MDS. Transplantation is a highly toxic treatment and given the advanced average age at diagnosis, only a minority of patients are eligible for curative treatment. Until the early 2000s, patients not eligible for transplant were mainly treated with frequent monitoring, supportive transfusion, growth factors, and iron chelation[13]. Since 2009, hypomethylating agents have improved the survival and quality of life of those with high risk MDS [13,14] and have become a mainstay of treatment. Treatments for MDS improve the quality of life of those living with the disorder but are associated with considerable cost[15]and toxicity[16]. Communicating the complexities of diagnosis, prognosis and treatment choices in MDS is an area that poses significant challenges to patients and clinicians. This is illustrated in a patient survey which showed that only 7% of patients with MDS had their condition described to them as a cancer[13]. Additionally, 55% of respondents did not know what prognostic category their disease fell into. Perhaps more concerningly, 31% of patients on supportive treatment alone believed their current treatment would prolong survival and 16% of all patients believed their treatment was curative, while 30% were uncertain of the effect of their treatment.

Despite advances in our understanding of the pathobiology of MDS and improvements in treatment in the last 15 years, the clinical care of patients with MDS remains largely focused on palliative treatment. Unfortunately, the research landscape does not reflect this reality. The genetic basis for MDS for example, is an area of intense research[17]. Over the same time, aspects such as transfusion optimization, iron chelation, health service delivery, end of life care and decision-making tools remain understudied and are often

extrapolated from other areas of hematology or oncology [18–20]. Moreover, despite repeated calls to choose meaningful research outcomes, the focus of most studies continues to be on morphologic or laboratory end points that are of little to no value to patients[21]. Thus, while our understanding of the pathobiology and prognosis of MDS has improved, our ability to improve the everyday experience of living with MDS has lagged.

Enabling people with MDS and the clinicians caring for them to participate in research prioritization on this condition would bring to the forefront important, clinically relevant questions in areas that are currently understudied. The expectation is that a greater focus on patient-centred research will translate into discoveries that will directly improve the lives of those living with this complex condition and their caregivers.

The James Lind Alliance:

In the United Kingdom, the National Institute for Health Research (NIHR) supports the James Lind Alliance (JLA) to act as a vehicle for patient engagement in research. The JLA is a non-profit initiative established in 2004 with the goal of ensuring that health research funders are aware of the issues that are most important to patients and clinicians. It engages patients, their caregivers and treating clinicians in a Priority Setting Partnership (PSP) to identify and prioritize research questions in a scientifically sound and transparent process. The result is a "top 10" list of research priorities where lack of evidence, termed "uncertainty" exists. The list is available in the public domain and can be used by researchers, advocacy groups, funders and policy makers alike to direct resources appropriately. This approach has been successfully used in the United Kingdom to spur observational studies and clinical trials that address treatment uncertainties in a variety of health care areas[22]. The value of ensuring research matches the needs of its intended users has been recognized by research funders such that the JLA infrastructure is funded by the National Institute for Health Research in the United Kingdom to oversee the process there. While the JLA approach has traditionally been used to identify areas of treatment uncertainty, it is also being used to identify gaps in

diagnosis, disease prevention and health care delivery [23,24]. In the Canadian context, there is no JLA parallel organization, but the JLA method has been used by Canadian groups to define research priorities in a variety of fields [25–28].

The James Lind Alliance Research Priority Setting Method:

The James Lind Alliance method to research priority setting is a five-step method consisting of : formation of a priority setting partnership, uncertainty gathering, data analysis, interim prioritization and final prioritization (Figure 1).

The cornerstone of the JLA method is the Priority Setting Partnership (PSP). This partnership is composed of clinicians, patients, caregivers and advocates working together as equals, to set priorities for research. Researchers who are non-clinicians are not included in the PSP. Classically, PSPs have tried to define uncertainties related to treatment of disease, but their scope has since expanded to include questions of health care team composition and service delivery [29]. This has led the JLA to update its terminology to the more inclusive "evidence uncertainty"[30]. In the setting of treatment uncertainties, uncertainty is defined as either a lack of systematic reviews addressing the question or that existing systematic reviews confirm the presence of uncertainty regarding effects of treatment. With the expansion of scope, however, this definition is not always relevant. Thus, each PSP is free to agree on criteria for determining uncertainties within its own scope of work. Once the steering committee is formed and agrees upon the scope and method of work, uncertainty gathering begins. This is usually done through surveys but can also be carried out through inviting relevant groups to submit research priorities directly without the use of a survey. Once the uncertainties (i.e. questions) have been gathered, they are collated and grouped using the methods agreed upon by the PSP. Next, the literature is searched to identify whether the questions have already been answered by the existing research. Only if the PSP considers the question not to have been answered, is it advanced through the next stages. The next step is for the list of questions to undergo interim prioritization by a broader group of stakeholders. Once again, exactly how this is done is left to the discretion of each

PSP. The interim priorities are then brought forward to a final workshop where they are discussed during a combination of break-out and plenary sessions, and a consensus list of top 10 priorities is agreed upon. The JLA guidebook[29] outlines possible methods for each step and important factors for groups to consider, but ultimately the most important requirement is that methods are transparently recorded and reported on. Although the final list of 'top 10' research priorities spells the end of the prioritization process, it is the beginning of the path towards re-aligning research with the needs of its end users.

To date, the JLA approach has been used by over 100 priority setting partnerships to define priorities in a diverse array of health conditions and professional settings[31]. Yet, despite its popularity, there is a paucity of data on its real-world implementation and its impact on research activity.

Structure of the Thesis:

This thesis is comprised of two manuscripts. The first is a scoping qualitative review of the barriers and facilitators faced by groups who have conducted JLA priority setting exercises in a variety of diseases. The second, is a report of our own experience conducting the first 2 steps of a priority setting partnership in MDS. Protocols were developed for each of the studies but were not published.

Thesis Objectives and Guiding Conceptual Framework:

The overarching goal of my thesis is to examine the implementation of the JLA approach as a method for public engagement in research prioritization. The specific objectives were to first, provide a comprehensive review of the barriers and facilitators faced during implementation, and second, to describe our own experience in implementing a PSP to prioritize research in MDS.

We are aware of at least one other scoping review of the JLA process[32], but it differs quite significantly from our review in its purpose, which is specifically focused on the barriers and facilitators, rather than providing an overall review of the methods used at each step.

Because I am focused on the JLA method (Fig 1), we used it as a guiding framework throughout the thesis. Our analysis is framed by the steps of the JLA method and delve into the methodology and the implications of our findings for the broader adoption of this method.

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Figures:

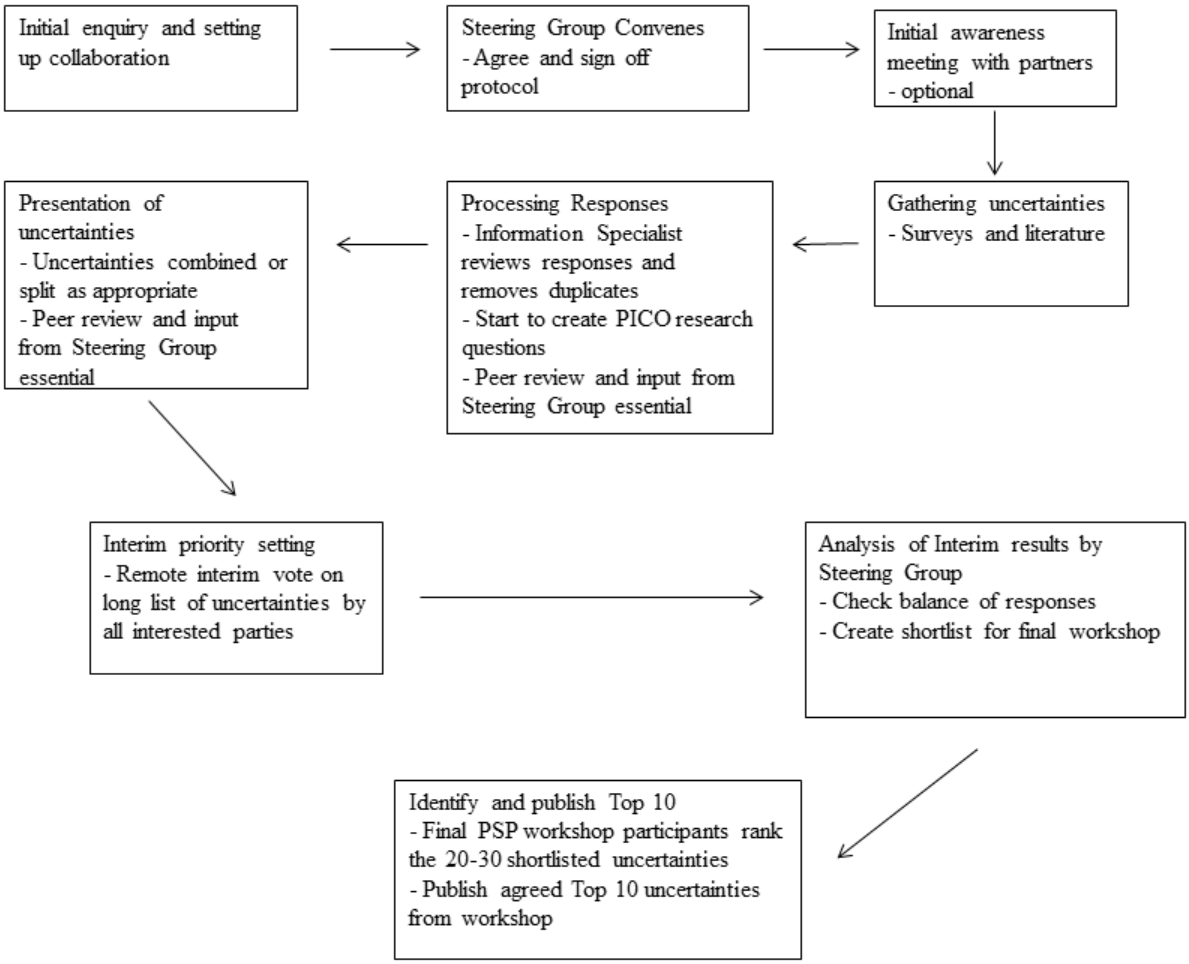

Figure 1: Overview of the JLA process – Adapted from the JLA Guidebook v6 (2016).

Chapter 2

Sixteen Years of James Lind Alliance Priority Setting Partnerships: A Scoping Review of Implementation Strengths and Challenges

This manuscript is formatted for BMJ Open with the exception of the word limit which is exceeded for the thesis version (not submitted)

Authors:

Grace Christou MD, MSc, FRCPC; Faculty of Medicine, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON, Canada

Natasha Kekre MD, MPH, FRCPC; Faculty of Medicine, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON, Canada

Risa Shorr MLS, Librarian, Learning Services, The Ottawa Hospital, Ottawa, ON

Dean Fergusson MHA, PhD, FCAHS; School of Epidemiology and Public Health, University of Ottawa, The Ottawa Hospital Research Institute, Ottawa, ON, Canada

Justin Presseau PhD; School of Epidemiology and Public Health, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON

Ian D Graham PhD, FCAHS, FNYAM, FRSC; School of Epidemiology and Public Health, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON

Statement of Contributions:

I designed and carried out the study in its entirety. Collaboration from co-authors was provided for literature search (Risa Shorr) and coding verification (Dr. Ian Graham).

Abstract:

Objectives: Involving patients, caregivers and front line healthcare professionals (HCPs) in setting research priorities has been recognized as an important step towards producing more impactful research. The James Lind Alliance (JLA) method has emerged as one of the main comprehensive methods for engaging these stakeholders in research priority setting. This study presents an in-depth analysis of the challenges and facilitators faced by groups who have implemented this process.

Design: A scoping review of studies using the JLA method for research priority setting to identify and describe barriers and facilitators encountered during the implementation of priority setting partnerships and categorize them into meaningful groups. We included articles on any health condition, medical intervention or field of study. Articles prioritizing research on entire professions or research methods were excluded.

Data Sources: MedLine search from 1974 to 2020.

Results: We included 68 articles published between 2009 and 2020. We categorized facilitators and barriers of the JLA method into 6 different categories corresponding roughly to stages of the 5 stages of the JLA method, plus a catchall "Other" category. Stakeholder engagement was the most commonly reported on category (64 articles), followed by final prioritization (31 articles), uncertainty gathering (23 articles) and collation and categorization of uncertainties (22 articles). Most of the reported data centred around diversity of stakeholders, data handling and usability of the results.

Conclusions: The JLA method is a comprehensive and valuable tool for broad public engagement in research priority setting. It enables inclusion of a broader range of voices and if applied thoughtfully, can yield very useful results. It can be a resource intensive process, particularly if the scope of the work is broad. Future implementation would benefit from the availability of additional tools to help facilitate interim prioritization.

Data Availability:

Raw data available upon request from corresponding author.

Strengths and Limitations of This Study:

- The first in depth analysis of the implementation of the JLA method across multiple health fields, worldwide.
- Data synthesized in a way to enable a bird's eye view of areas where additional planning may be required to aid future implementation.

- Analysis limited by the lack of detailed descriptions of the challenges and how these were overcome in most articles.
- Analysis is based on authors' perception and not independently assessed.

Introduction:

Several studies have shown a misalignment between areas of intense research activity and the priorities of people living with health conditions, their caregivers and front-line medical practitioners[2,3]. A recent analysis comparing patient and clinician priorities across 14 health conditions with ongoing research in these areas demonstrated that while drugs constituted just 19% of patient and clinician priorities, they accounted for 37% of non-commercial trials and 89% of commercial trials[4]. This mismatch contributes to diminished efficiency of research where areas of great clinical need remain unstudied, while certain areas of low clinical relevance have high research activity. This leads to diminished research utilization and a perception that academic science does not serve the public interest. To address this imbalance, research funders have encouraged or required greater involvement of patients in research to ensure their voice is included in decisions relating to research funding, design, implementation and evaluation (Canada's Strategy for Patient Oriented Research, UK Involve, Patient-Centred Outcomes Research Institute)[33–35].

Involving patients and caregivers in identifying research priorities is important for several reasons. First, patients possess a unique perspective due to their lived experience with a condition and their particular social context[7]. Including their voice can uncover important research gaps that would otherwise remain unknown. Second, meaningfully engaging the public leads to improved communication and greater mutual trust and respect between researchers and the public[6]. The cumulative effect is that research is more transparent and accountable and more likely to answer clinically relevant questions, making it more useful to its end users[36]. The James Lind Alliance (JLA) was established in 2004 in the United Kingdom as a non-profit organization with the expressed goal of promoting patient and clinician involvement in research prioritization, thus ensuring that future research has greater impact on patients' lives by addressing areas of

greatest importance to them[37]. The organization provides the infrastructure and support needed to bring together the various stakeholders as equal partners in priority setting partnerships (PSPs).

Since its inception, the JLA organization has conducted almost one hundred PSPs where patients, health advocacy groups, health care professional associations and individual clinicians all work towards an agreed upon list of top 10 research priorities. Several groups have conducted PSPs with JLA support while others have applied the JLA approach independently. The 5 step JLA method was initially developed to prioritize uncertainties related to treatment effects that merit further research, but has been applied to a much broader scope of questions including diagnosis, prognosis, treatment and care delivery[29]. Despite the attraction and growing popularity of this method, there has not been a published analysis of the real world experience of its implementation, commonly encountered pitfalls and how various groups have overcome them. To inform future iterations of this process and enhance the utility of these multi-stakeholder partnerships, we conducted a systematic review of barriers and facilitators to the work of PSPs.

Methods:

We conducted a scoping review using the Joanna Briggs Institute (JBI) scoping review framework revised by Peters et al, 2020[38]. A protocol was developed apriori and closely followed in keeping with best practice. Here we report our results using the PRISMA-ScR checklist[39] .

Eligibility Criteria:

We included all full-length original articles using the JLA method[30] to priority setting partnerships that had a goal of setting research priorities in any health condition regardless of whether they directly involved the JLA organization. Eligible articles were those that reported on the work of PSPs and followed all basic steps of forming a PSP, gathering uncertainties, verifying uncertainties, interim prioritization and final prioritization as outlined in the JLA guidebook. Articles from PSPs that followed all steps were included regardless of any adaptations made. We included articles if they reported on at least one strength or weakness of the work. We excluded conference proceedings, review articles, general commentaries,

editorials, and articles that omitted steps of the JLA method or those that set priorities for studying research methods (e.g. clinical trials) or entire professions (e.g. physiotherapy).

Outcomes:

The primary outcomes of the study were descriptive barriers and facilitators to using the JLA method for research priority setting as described in the source articles. There were no secondary outcomes.

Literature Search:

We initially searched MedLine from 1947- February 2019 for eligible articles in English or French using a broad search strategy designed in collaboration with an information specialist at our institution. The search strategy underwent PRESS review to ensure comprehensiveness[40]. The complete search strategy is shown in Appendix 1. We also manually searched the JLA website for any PSPs that were not identified by our literature search and conducted a specific search using PubMed and Google Scholar to locate any eligible articles from these PSPs. In October 2020, a search of PubMed was conducted using only "James Lind Alliance" as a search term for articles published between January 1st, 2019 and October 7th, 2020.

Identification of studies:

Search results were imported into Mendeley™ and duplicates removed. Titles and abstracts were screened and included even if it was unclear that the JLA method was used. Full length articles were then screened to ensure selected records addressed a health condition, intervention or field, followed all steps of the JLA method and reported on barriers and facilitators.

Data Extraction:

For each included record we collected the health condition or health intervention, year of publication, date of PSP's work, extent of JLA involvement as a categorical variable, geographical region covered by the PSP to the extent possible, and any facilitators and barriers reported by the manuscript authors. Data was extracted by a single reviewer (GC).

Data Analysis:

Qualitative data were analyzed inductively[41] i.e., one where there were no pre-defined codes or themes at the start of the analysis. First, raw data was extracted and entered into an Excel sheet. It was then coded using preliminary codes that emerged organically. Codes were refined using the constant comparative method[42] as more codes emerged and relationships between them became clearer. Initial coding was undertaken by one author (GC) and verified independently using a list of codes by a second author (IG). Discrepancies were resolved by consensus. Coded data were collated into topics, allowing the identification of broader themes that emerged from the data.

To organize the collected qualitative data, we adapted the approach taken by the pressure ulcer PSP in their report [43]. Data were divided into 6 broad categories roughly corresponding to the stages of implementing the JLA method. These are stakeholder engagement, uncertainty gathering, collation and categorization of uncertainties, interim prioritization, final prioritization, and a catchall "other" category.

Results:

Literature Search:

Together, our searches yielded 2329 citations, 2136 of which were excluded by duplicate removal and titles and abstract screening. A further 128 were excluded at full text screening, leaving 65 full text articles (Figure 1). A hand search of the JLA website together with a targeted Google and PubMed search identified 3 more articles bringing the number of eligible articles to 68.

Study Characteristics:

Articles were published between 2009 and 2020 with 60 (88%) published between 2014 and 2020 (Table 1). Fifty were conducted within the United Kingdom, 11 in Canada, 2 in Spain, and 1 each in Australia and Sweden. A further 3 were international in scope. The JLA was explicitly involved in 56 (82%) of PSPs,

either in conducting the exercise or in lending its expertise in a specific capacity. It should be noted that methods for reporting the JLA's role were not uniform across articles.

PSPs addressed research priorities related to specific health conditions such as hypertension or type I diabetes (50), medical interventions like the use of digital technology in mental health (7) or entire fields of care like anesthesia and critical care (11). In 3 instances, separate journal articles reported on work undertaken as part of a larger PSP or used the results of a related PSP (FREE TEA, Alopecia Areata and Aphasia). In the case of the 2 articles from the alopecia PSP, there was complete overlap in the discussion of strengths and limitations. Therefore, we decided to exclude the later article from further analysis[44]. In the case of the FREE TEA article[45], there was sufficient difference in methods and scope to treat it as a separate article, bringing the total number of articles to 67 articles reporting on the work of 66 distinct PSPs.

Barriers and Facilitators:

Our analysis uncovered the following barriers and facilitators reported by groups conducting PSPs.

1- Stakeholder Engagement: (64 articles, table 2)

i. ***Factors related to the health condition/intervention:*** Isolation resulting from the nature of the condition of the affected population was a barrier to effective stakeholder engagement in 7 PSPs on dementia, Parkinson's disease, mesothelioma, pressure ulcer, kidney disease nearing hemodialysis, gestational diabetes and fragility fractures, whereas stigma was identified as a barrier to engaging with people affected by alcohol related liver disease and frailty. In contrast, the head and neck cancer PSP felt this approach was successful at engaging with a normally isolated population and identified it as a strength of their study. In the Epidermolysis Bullosa (EB) PSP, disease rarity contributed to difficulty in finding adequate numbers of patients, yet because of the rare nature of the disease, most patients with the condition were connected to advocacy groups and could be easily found. Furthermore, the rarity of the condition enabled the PSP to identify experts in the field by reaching out to authors of scientific papers.

Disease burden was identified as a barrier to engagement with people living with severe dementia, type 2 diabetes, fragility fractures and rare musculoskeletal diseases. Caregivers of people who had suffered stroke (2 articles), spinal cord injury, or were admitted to the intensive care unit were also difficult to engage due to the high emotional toll and care requirements associated with these conditions. For young people with cancer, there was low participation from those undergoing active treatment.

A lack of well-defined and cohesive patient advocacy groups accounted for difficulty connecting with the affected population in 5 articles dealing with pressure ulcers, emergency medicine, alcohol related liver disease, blood donation and malnutrition. The fibromyalgia and frailty PSPs felt that engagement was made more difficult by the lack of specific diagnostic criteria defining these conditions.

ii. ***Stakeholder Diversity:*** This was the most commonly reported topic with 40 articles reporting either a strength or a weakness. Twenty PSPs felt that ethnic minorities were underrepresented whereas PSPs on miscarriage and type 1 diabetes felt that their methods specifically targeting ethnic minorities were successful. Six studies raised a concern about potential lack of engagement with people from disadvantaged socioeconomic groups and none of them thought that this problem had been addressed adequately using usual methods. Thirteen articles reported on geographic representation with 6 studies finding it uneven and 2 reporting a mixed outcome. Engagement with different genders and sexual minorities was reported in 9 articles. The PSPs on acne, lichen sclerosus, psoriasis, frailty, young people with cancer and rare musculoskeletal diseases found that there was under representation of men in their sample, while the primary care PSP had inadequate LGBTQ2 representation. Eleven studies reported difficulties recruiting people from the full age range affected by the condition, while one study on learning difficulties in children engaged a high proportion of young people. In the PSP on endometrial cancer, it was recognized early on that most survey respondents were younger women whose concerns differed significantly from more frequently affected older women. To overcome this age imbalance, older women were specifically invited

to take part in the interim prioritization and final workshop. In contrast, the pressure ulcer PSP felt that younger patients with pressure ulcers were significantly underrepresented in their sample. Six PSPs were concerned that most participants were already accessing healthcare and advocacy groups, leaving people with poor access to care, underrepresented.

iii. ***Healthcare provider (HCP) Participation:*** Overall, 8 articles reported on the extent of HCP participation, but only 2 (type 2 diabetes and tinnitus) identified good HCP participation as a strength. The prostate cancer PSP in particular, recorded very low participation by oncologists and urologists. Other PSPs that had difficulties in this regard included fibromyalgia, pessary use, childhood disability, primary care and rare musculoskeletal diseases.

iv. ***Civil Society Context:*** The strength of patient advocacy organizations concerned with the target health condition was an important factor discussed by 9 articles. The presence of a strong organization was a strength to both of vitiligo and cystic fibrosis PSPs. In contrast, the pressure ulcer PSP suffered from the lack of a strong advocacy group to partner with, given that ulcers can occur as a complication of many other conditions. For the acne PSP, the presence of a strong advocacy group for polycystic ovary syndrome was helpful in garnering high participation, but created an imbalance whereby the group with the highest participation was not representative of those who live with acne in general. A lack of participation from advocacy groups was noted as a weakness by the PSPs on alopecia, childhood disability, emergency medicine, alcohol-related liver disease and acne. The pressure ulcer PSP noted that in addition to the lack of an advocacy group, there was very little non-industry led research in the field.

v. ***Mechanism of Engagement:*** Twenty articles reported on the form and function of the steering committee as either a facilitator or a barrier. The pressure ulcer PSP cited an effective introductory meeting during which the steering committee was recruited as an important facilitator. Additionally, the steering group was given significant autonomy to decide on the mechanism of engagement with the broader public

and uncertainty gathering. A similar hands-off approach to engagement was considered a strength of the urinary incontinence PSP. Ten articles cited broad stakeholder involvement as a strength, although in-depth analysis of this aspect was lacking. One article on idiopathic intracranial hypertension reported that although the diversity of stakeholders strengthened the work overall, it posed significant challenges during prioritization steps. The dementia, blood transfusion and type I diabetes PSPs cited a collaborative spirit between various stakeholder groups as an important strength of their work. The use of web-based meeting software was a vital tool for the cystic fibrosis PSP as people affected with this condition would not otherwise be able to meet due concerns over bacterial cross-contamination between people. Advertising was discussed by 9 articles. The lack of social media access was a significant barrier to the pressure ulcer and asthma PSPs. Additionally, 2 PSPs took advantage of advertising through traditional channels and found it an effective engagement strategy. Specific targeting of special populations was successfully undertaken by the PSPs on type 2 diabetes, EB, endometrial cancer and aphasia after stroke.

Twenty-three articles discussed survey administration in their list of strengths and weaknesses. The use of an online survey was seen as a strength by 3 PSPs, a weakness by 8, and received mixed reviews by 2 others. Four PSPs (digital technology in mental health, tobacco control and smoking cessation, life after stroke, and alopecia) considered face-to-face meetings to gather uncertainties as a strength of their approach. PSPs on ADHD and aphasia after stroke used multimodal communication methods to gather uncertainties while the dementia PSP encouraged group responses. All 3 felt that these approaches facilitated better participation from populations facing communication challenges. A fourth PSP on advanced heart failure used a simple language survey to simplify data collection. The PSPs on alcohol-related liver disease and fragility fractures found involving HCPs in directly administering surveys was an important means of engaging otherwise isolated patient populations. Two studies reported on incentives to encourage survey uptake, with mixed results. The acne PSP noted that while the use of incentives encouraged greater number of submissions, many of these were submitted blank. On the other hand, the

tobacco control and smoking cessation PSP felt that the use of incentive vouchers improved survey completion.

Summary: Several factors affecting the ability to engage with diverse stakeholders were identified. These included the particular effects of the health condition itself on people's ability to participate in research prioritization, whether strong disease advocacy groups with wide reach could be identified and recruited and to what extent HCPs were mobilized to participate. Many articles expressed concerns regarding the diversity of participating voices, particularly as it pertains to those disadvantaged by economic, educational, linguistic or ethnic barriers. These concerns are directly linked to the ways in which partnerships were built, advertised and surveys disseminated. A major theme is that although social media and electronic surveys facilitated the work overall, over-reliance on these methods may contribute to the exclusion of marginalized groups. Facilitators within this area included using methods tailored specifically to the target population.

2- Uncertainty Gathering: (23 articles, table 3).

i. ***Over and under response to surveys:*** The PSP on urinary incontinence cited lack of funding as the main reason for relying on partner organizations to devise their own methods for gathering and prioritizing uncertainties, thus making it difficult to assess validity of methods. In terms of survey response, 3 articles on pressure ulcer, vitiligo and retinoblastoma noted poor survey uptake. On the other hand, the tinnitus PSP had a very high response to its survey which led to difficulties with data handling in subsequent stages. Concerning the quality of surveys submitted, the articles on head and neck cancers and acne PSPs both reported a high percentage of blank surveys being submitted and a number of difficulties with the quality of responses was noted in other articles.

ii. ***Formulation of research questions:*** Three articles on pressure ulcers, tinnitus and asthma noted that the public may have difficulty with the concept of an "uncertainty". The pressure ulcer PSP article felt

that the term was stigmatizing and may serve to degrade public trust in current medical care. This was overcome in part by explaining the term and framing survey questions positively. Several articles noted that the public had difficulty understanding what research can achieve and formulating suitable questions. Questions were often submitted as personal anecdotes as noted by the pressure ulcer, alopecia, Parkinson's disease, spinal cord injury and acne groups, among others. The pressure ulcer, cardiac surgery and hyperacusis PSPs in particular were able to provide a facilitator to reframe complaints into potential research questions. In contrast, the Parkinson's disease PSP did not have access to this resource and its article noted that a number of submissions raised concerns about the physical or psychological safety of the respondent with no way to follow-up. In the retinoblastoma PSP, researchers applied the PICO formula consistently to question formulation which authors of that article noted as a strength.

iii. ***Scope of PSP:*** The diversity of questions submitted by HCPs and people affected by the condition was also discussed. The vitiligo PSP noted as problematic that broad questions were submitted by patients whereas very specific questions were submitted by healthcare providers and researchers. In the epilepsy article, there was concordance between HCP and patient questions. The scope of submitted questions was considered inappropriate in 3 articles. In the main life after stroke and primary care articles, the questions submitted were considered too broad, limiting their utility. Whereas the article on the childhood disability noted that narrowing the scope of questions too early in the process was a weakness of their approach.

iv. ***Source of uncertainties:*** One group dealing with aphasia after stroke used uncertainties from the life after stroke PSP but added more from the final prioritization workshop and considered it a strength. The hypertension PSP included questions that had been answered by research and found this a weakness.

Summary: Gathering potential research questions from the public was complicated by several factors including under or over response to surveys, submission of a large number of blank surveys, research topics being buried within anecdotes and a lack of public familiarity with the process of scientific research and

question formulation. Some groups overcame these barriers by investing in resources to educate potential respondents and reformulating anecdotal submissions into appropriate research questions. The scope of PSPs impacts the volume and diversity of responses submitted and various groups identified the mismatch between scope of work and the resources available as problematic.

3- Collation and Categorization of Uncertainties: (22 articles, table 4)

Barriers and facilitators during this stage of implementing the JLA method mainly concerned data handling, question formulation and literature appraisal (table 4).

i. ***Data Handling:*** Six articles discussed data handling. The tinnitus and alopecia groups had a very large number of questions submitted. The shoulder surgery and epilepsy PSPs noted that collating and categorizing the data was extremely resource intensive. Additionally, 4 PSP's (tobacco control, tinnitus, alopecia, gestational diabetes) had difficulty categorizing submitted questions.

ii. ***Question Formulation:*** Fifteen articles noted barriers and facilitators in this topic. Four articles on pressure ulcers, shoulder surgery, stillbirth and knee arthroplasty felt that the tension between broad and specific questions was a challenge of the JLA method. There was no consensus on the optimal means of dealing with this tension. PSPs on Parkinson's disease, fragility fractures and sight and vision loss considered their ability to retain granular question detail as a strength, while 3 other PSPs on anesthesia, acne and lichen sclerosus chose to collate specific questions into broader ones and considered that a strength. Groups prioritizing research on mesothelioma and childhood disability noted difficulty formulating questions from submissions, with the latter group opting to remove questions if they could not be properly formulated into ones answerable by research. The vitiligo group circumvented potential problems with question formulation by applying a standard format to all questions and felt that this significantly streamlined the process.

iii. ***Literature appraisal:*** Six PSPs discussed their approach to literature assessment as either a weakness or a strength. In this area, the term "indicative uncertainty" made several appearances. "Uncertainty" is the term originally used by the JLA organization to describe an area of research where the effect of treatment was still unknown or "uncertain". It is roughly equivalent to the concept of clinical equipoise. An indicative uncertainty is an umbrella question under which more specific related questions can be grouped. When conducting a literature search, only the broader question is examined, making literature appraisal less intensive. In the articles on pressure ulcers, type 2 diabetes and primary care, the use of indicative uncertainties was noted as a facilitator in assessing the literature. The epilepsy PSP chose to only assess the literature surrounding the top 60 of 180 uncertainties. While this cut down on the resources needed, the article did note this as a potential limitation of their work. The urinary incontinence PSP reported general difficulty assessing the literature, while the miscarriage PSP had difficulty adapting the original JLA concept of "treatment uncertainty" to a broader scope of questions. The pressure ulcer PSP found the expertise of an information specialist in searching and appraising the literature beneficial.

Summary: Analysis of large volumes of qualitative data is a resource intensive step in the conduct of PSPs and one that was challenging in many cases. Questions around one topic can be heterogeneous in focus and breadth, making precise classification difficult. One popular approach for overcoming this challenge is the adoption of an aggregative method of analysis. Using this approach, all questions pertaining to a broad topic are filed under an "indicative uncertainty" which becomes the subject of a literature search. The involvement of an information specialist during this phase is an important facilitator to preserve result validity.

4- Interim Prioritization: (12 articles, table 5)

i. ***Tools for interim prioritization:*** Two articles noted challenges in finding appropriate tools for interim prioritization. Participants in the pressure ulcer PSP had difficulty understanding the concept of

prioritization, but the group overcame this by using a simple prioritization scale and creating a YouTube® video demonstrating its use. Meanwhile, the tobacco control group had similar difficulties, but lacked the resources for tool development.

ii. ***Fairness/Equity:*** Ten articles reported on the fairness of the prioritization. Four articles (digital technology in mental health, life after stroke, congenital ichthyosis and fragility fractures) considered it a weakness that broad questions were assigned a higher priority than more specific ones. In the stillbirth PSP, frequency-based prioritization led to important "taboo" questions like domestic violence and drug misuse being excluded from the final discussion. In the article on mesothelioma, differences in the interim priorities of stakeholder groups were difficult to reconcile. Three PSPs on emergency medicine, type 2 diabetes and hyperacusis actively tried to achieve a fair list of priorities, but each used a slightly different approach. In the type 2 diabetes PSP, the concerns of ethnic minorities were specifically included in interim priorities. The emergency medicine PSP group attempted to balance priorities of various subspecialties, considered the prevalence of conditions and whether questions were covered by other PSPs. The hyperacusis PSP used a weighted ranking to ensure that questions submitted by people with lived experience were prioritized.

Summary: The main issues reported during the interim prioritization phase centred on the lack of standardized tools and methods for prioritization. Broader questions with wider appeal were reported to generally be given higher priority than more focused questions. Certain PSPs were concerned with the fairness of the prioritization process and used various methods to ensure the diversity of questions carried forward during interim prioritization. Issues of fairness and representation at this stage relate directly to those discussed during the stakeholder engagement phase.

5- Final Prioritization: (31 articles, table 6)

Strengths and weaknesses concerned attendance, discussion, record keeping and final prioritization.

i. ***Workshop Attendance:*** Fourteen articles reported on attendance at the final workshop. Four noted difficulties securing attendees. The pressure ulcer PSP had difficulty getting nursing home residents affected by the condition to attend the workshop due to "administrative barriers", while the tobacco control PSP suffered a lack of HCP participation. The kidney transplant PSP had 20 attendees, falling short of the optimal 25, but recorded strengths in other aspects of the final prioritization. Three articles cited strengths in the representativeness of attendees. The kidney transplant, intensive care and sight and visions groups felt that there was equal representation of HCPs and patients. The Canadian Anesthesia PSP had more patients and caregivers at the final workshop which balanced out a higher HCP response to the survey. The PSPs on fragility fractures, retinoblastoma, hyperacusis and human milk banking on the other hand, felt that attendance at the workshop may not have been representative of the affected population. Adapting the design of the final workshop facilitated workshop participation for the hyperacusis, dementia, cystic fibrosis (CF) and EB partnerships. Web-based meeting technology was important to allowing the meetings on CF and EB to proceed, while the dementia group adopted a dementia-friendly workshop design.

ii. ***Group Discussion:*** Twenty-two articles reported on various aspects of the discussion with 9 articles citing an open discussion where views were freely and enthusiastically exchanged as a strength. In the ICU article in particular, authors noted that attendees were able to bring a broader perspective to the table, extending the dialogue to include the views of those unable to attend. In contrast, 5 articles on tobacco control, congenital ichthyosis, ADHD, mesothelioma and idiopathic intracranial hypertension found the discussions challenging, identifying a variety of factors like lay people being disadvantaged, domination of the discussion by researchers and potential bias of the participants as factors. The article on type I diabetes noted the balance between larger plenary sessions and smaller group sessions as an important facilitator to a productive discussion. The expertise of JLA facilitators was valuable in facilitating the discussion and ensuring equal participation in 7 articles (tobacco control, type I DM, kidney disease nearing dialysis, EB, healthcare-associated infections and pregnancy hypertension).

iii. ***Record keeping:*** The tobacco control and smoking cessation PSP had 2 specific difficulties with record keeping. The recordings of the discussions had poor sound quality and were of limited use and there was poor tracking of participation and voting, making it difficult to evaluate the results.

iv. ***Top 10 Selection:*** Twelve articles discussed the selection of the "top 10" priorities. Authors of the mesothelioma article felt that the final list of priorities had moved away from patient-centred questions. Both the aphasia after stroke and the life after stroke PSPs chose to change the scope of prioritized questions. The groups' analysis of the effect of this adaptation differed, however. The article on aphasia after stroke considered this a positive demonstration of flexibility and responsiveness to stakeholders, whereas the life after stroke article considered it a weakness which undermined methodologic rigor. Six articles noted that only broad questions were included in the final 'top 10" priorities. Some, such as the childhood disability and alopecia PSPs viewed this as a weakness as these questions were less likely to be answerable by research, whereas the EB group, for example, found it a strength in that broader priorities applied to the greatest number of affected people. Three PSPs (head and neck cancers, alopecia and endometrial cancer) were able to achieve consensus, while 2 PSPs (mesothelioma and hypertension) found this difficult.

Summary: Articles highlighted strengths and limitations to the final prioritization phase. Attendance at the final prioritization workshop was variable among PSPs with under representation on healthcare providers and those severely affected by the conditions of interest being the most prominent weaknesses. In a few cases, attendees were able to effectively represent the perspectives of those not physically present, which enriched the discussion. A balanced mix of small group and large plenary discussions facilitated by trained JLA personnel were cited as advantageous. This format allowed for inclusive and productive discussions and avoided having one group dominate the proceedings. One important issue highlighted is the ongoing tension between broad and specific questions. It was noted that broad questions will need further refinement

into focused questions that may be more answerable by research. Finally, partly because of this ongoing tension, it was not always possible to achieve consensus among stakeholder groups on the "Top 10".

6- Other:

In addition to the specific issues highlighted above, concerns about the generalizability of the findings was cited in 8 articles, others alluded to the issue indirectly. Some articles also highlighted that assessing the effectiveness of stakeholder engagement is limited by the inability to calculate a response rate and at times, by the lack of demographic data collection.

Discussion:

There has been growing interest in engaging patients and frontline clinicians in setting research priorities in an attempt to address waste in clinical research and improve the uptake of scientific findings. The JLA approach to priority setting has been adopted by various groups to prioritize an ever-growing scope of research questions. With just over 100 PSPs convened, we set out to review the challenges faced by these groups and the ways in which they were circumvented, with the goal of improving the utility of this approach to research prioritization.

Our review reports on findings of 68 published articles of 67 PSPs, all of which discussed at least one challenge and/or strength of their work using the JLA method. Notably, while the UK was the leading site for implementation of PSPs, Canada was the second most common site. A key finding of our review is that most articles did not report on the strengths and limitations of their experience in any detail. This limits our ability to conduct a full in-depth assessment of the JLA method. Nonetheless, we were able to glean some important insights.

Most of the reported challenges can be classified into 3 overarching themes: representation, data handling and usability of results.

Representation: Effective engagement was partly dependent on forming an effective partnership with professional organizations and advocacy groups representing the full spectrum of stakeholders. Identifying

and partnering with these organizations loosely correlated with the breadth of the health research topic. For health conditions affecting a well-defined target population, identifying key stakeholder organizations and engaging with them is likely to be a simpler and more streamlined process. On the other hand, health conditions that have several potential underlying causes (e.g. acne, pressure ulcer) or encompass broad fields of inquiry (critical care, primary care) require more extensive stakeholder analysis and a greater degree of coordination among groups. In turn, this may dictate a mixed method approach to engagement, uncertainty collection and prioritization.

Although many of the reported concerns centred around difficulty reaching isolated and disadvantaged populations, most PSPs did not collect or report detailed demographic information. Moreover, there are no clear criteria or widely accepted tools for evaluating engagement[46,47] and each group must set out its own evaluation framework and process. It is unclear what criteria were used and to what extent these gaps in representation threaten the validity of findings.

Engagement with marginalized populations requires specific planning and can be time and resource intensive for both researchers and the target population[48] and may not be required for valid results in every case. For some conditions or fields of study, it may be sufficient to capture a diversity of views and present the results of the PSPs along with detailed demographic information such that findings can be contextualized. In situations, where equity seeking populations are disproportionately affected by a condition, it is necessary to invest the resources needed to meaningfully engage with this hard to reach group, either as part of a broader JLA PSP or a more targeted approach to priority setting using best practice[49,50]. Even if deep engagement with underserved populations is beyond the reach of most PSPs, there are several areas that can be improved upon to promote greater diversity of participation.

The use of the internet and social media is a valuable facilitator to this type of public-facing work, but over-reliance on the internet may contribute to the continued exclusion of marginalized populations that have lower rates of internet use and political engagement online[51–54]. Additionally, the language in which surveys are administered likely impacts the ability of indigenous peoples and recent immigrants to participate. Thus,

making surveys available in several languages and formats would go a long way towards encouraging broader participation.

The vast majority of groups used random sampling, where a survey was launched and advertised through partner organizations. Using sampling techniques such as purposive sampling, where populations are defined based on particular criteria and specifically targeted, and snowball sampling, where participants with whom contact has been made recruit others in their group[55], would improve the diversity. We found that involving HCPs in survey administration, conducting face to face meetings with target groups, encouraging group responses and providing alternative means of communication were all successful in various partnerships.

Data Collection and Handling: Many methodological barriers were identified within this section. A few groups identified how they were able to overcome them, as discussed within the results section above. The collection of uncertainties itself was identified as a challenge by several groups. One of the main barriers to this work is that scientists communicate using logical-scientific communication which is centred on facts regardless of context, whereas "non-expert" populations communicate using narrative[56]. Thus, when many of the responses received were in the form of personal anecdotes rather than research questions, this was seen as a challenge to data collection. An alternative view is that these anecdotes contain a wealth of information about the unsolved issues people are facing with their condition. Seen from that perspective, consistently mining anecdotes for potential research questions is an important means of democratizing participation and deriving a fuller benefit from the resources expended in administering and answering surveys.

The JLA method was originally developed to prioritize uncertainties around the effect of treatments[30] (JLA Handbook), but the scope of PSPs has steadily grown and often includes any questions about any aspect of care. This has contributed to greater volumes of heterogenous qualitative data being collected and increasing demand on resources for data processing. Any groups considering this approach should have a plan for the handling of large volumes of qualitative data including a clear process for data coding,

categorization and conflict resolution. An exhaustive literature search and appraisal was equally demanding. Several PSPs included in our review, attempted to streamline the process through the use of indicative uncertainties around which literature searches are centred. The involvement of an information specialist who can develop a pragmatic approach to literature searches is indispensable.

In terms of literature appraisal, the original JLA method considers a question unanswered by research if there are no systematic reviews to answer it or if systematic reviews conclude that there is uncertainty around the effect of treatments[30]. This strict definition is difficult to apply to a broader scope of questions, thus one way to make PSPs more manageable is to re-narrow their focus on specific aspects of care.

Result Usability: The group discussions at the final workshop were generally viewed favourably. A recurrent theme is that broader topics were ranked higher than more specific research questions, presumably because a consensus-based process favours topics with broader appeal. The fact that broad topics reached the final consensus is likely a result of the ever-broadening scope of PSPs. It would be interesting to compare the uptake by researchers of general vs specific priorities and look at whether continuing to use a consensus-driven process for final prioritization is yielding useable results in the context of PSPs with a broad scope.

Strengths and Limitations: Although there is a published review of all JLA priority setting partnerships[32], the focus of that review was on cataloging the PSPs conducted thus far. To the best of our knowledge, our review represents the only in-depth, systematic analysis of the experiences of those who have used the JLA method. We present a synthesis of the common challenges that emerged and some of the adaptations that have yielded positive results. Our work was limited by the published record, which often focused on reporting the Top 10 research priorities, rather than a detailed account of the process. Our analysis would have been enhanced by directly communicating with all groups that have conducted PSPs and collecting

primary data specifically surrounding barriers and facilitators to the work, but this was not done due to time and resource limitations.

Implications for The JLA method as a vehicle for research priority setting: Overall, the JLA method provides a framework for a comprehensive approach to health research prioritization that gives voice to people affected by the condition and their health care teams. Based on our findings, it is not a method that is easily adapted for deep engagement with minority or equity deserving groups. Furthermore, its implementation requires attention to details unique to each condition, intervention or field of study. Perhaps it is not surprising that the majority of completed PSPs actually involve the JLA itself with its considerable resources and expertise.

Our analysis shows that the JLA priority setting process has worked best at prioritizing research in clearly defined health conditions with well-established stakeholder organizations as partners. It is primarily designed to understand the research priorities of the general population and if implemented thoughtfully, may be able to capture a diversity of voices.

Furthermore, the scope of research questions to be prioritized may have a direct impact on both the volume and specificity of responses collected and potentially on the nature and usability of the "Top 10" priorities. Additionally, criteria for evidence appraisal differs based on the scope of inquiry. Clinical trials and systematic reviews were used in the original JLA method to appraise literature around effects of treatment, but whether they are an appropriate benchmark for evaluating data about diagnostics and health care delivery remains an open question.

Finally, the methods for interim and final prioritization should be carefully thought through by each group undertaking PSPs. Methods for interim prioritization varied widely and each of these approaches has its own strengths and weaknesses and the selection should be guided by the goals of the exercise itself. The final prioritization is one largely driven by consensus-building. In several PSPs, general questions with broader appeal were ranked higher than more specific questions, leading some groups to question the utility

of the results. There too, the criteria for final prioritization should be set based on the specific goals of the exercise and should be acknowledged as such. Finally, it is unlikely that the JLA method, or any other method, will result in a universally relevant set of research priorities in any field. Many of the challenges reported reflect this reality, but in our view, do not detract from the overall utility of the JLA method as an important conduit for the voices of people affected by health conditions and those who care for them.

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Tables and Figures:

Figure 1 – Scoping Review PRISMA flow diagram

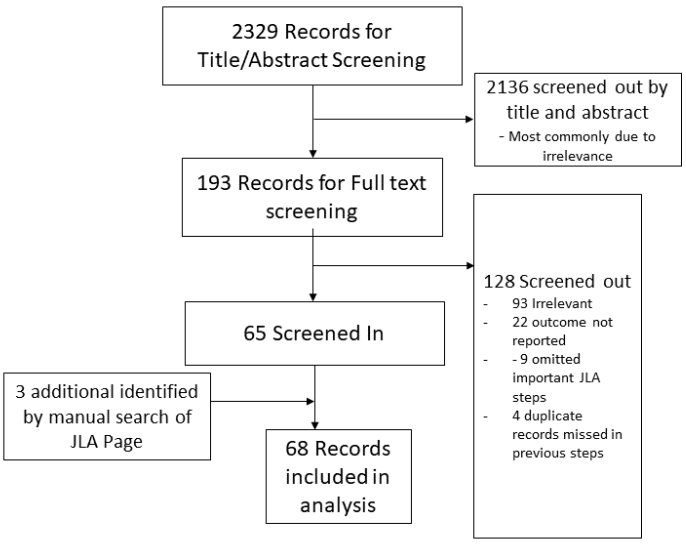

Table 1: Included Study Demographics

Health Condition/ Field of Study	Year Published	Country	JLA Involved	JLA Role
Epilepsy[57]	2010	UK - Wales	Yes	Conducted by the JLA
Asthma[58]	2010	UK	Yes	Conducted by the JLA
Urinary Incontinence[59]	2010	UK	Yes	Conducted by the JLA
Prostate Cancer[60]	2011	UK	Yes	Conducted by the JLA
Vitiligo[61]	2011	UK	Yes	Convened by the JLA
Type 1 DM[62]	2012	UK	Yes	Facilitator, possibly other
Dystrophic Epidermolysis Bullosa[63]	2013	Spain	No	None
Tinnitus[64]	2013	UK	Yes	Conducted by the JLA
Hidradenitis Suppurativa[65]	2014	UK	Yes	Unclear
Intensive Care Research[66]	2014	UK	Yes	Steering committee chair and meeting facilitator
Life after Stroke - Main[67]	2014	UK	Yes	Conducted by the JLA
Life after Stroke - FREE TEA[45]	2014	UK - Scotland	Yes	Unclear
On or Nearing Dialysis[68]	2014	Canada	Yes	Advisor and facilitator
Parkinson's Disease[69]	2014	UK	Yes	Facilitator
Sight Loss and Vision[70]	2014	UK	Yes	Steering committee chair and meeting facilitator
Acne[71]	2015	UK	Yes	Conducted by the JLA
Anesthesia and Peri-op care[72]	2015	UK	yes	Advisor
Childhood disability[73]	2015	UK	Yes	Chair
Congenital Ichthyosis[74]	2015	Spain	Unclear	Unclear
Mesothelioma[75]	2015	UK	Yes	Steering committee chair and meeting facilitator
Spinal Cord Injury[76]	2015	UK	Yes	Support and guidance

Stillbirth[77]	2015	UK	Yes	Steering committee chair and meeting facilitator
Alcohol-related Liver Disease[78]	2016	UK	Yes	Conducted by the JLA
Endometrial Cancer[79]	2016	UK	Yes	Advisor and facilitator
Gestational Diabetes Mellitus[27]	2016	Canada	No	None
Kidney Transplant[80]	2016	UK	Yes	JLA Chair
Pressure Ulcer[43]	2016	UK	Yes	Steering committee chair
Shoulder Surgery[81]	2016	UK	Yes	Advisor, co-ordinator, data analyst
Alopecia Areata[82]	2017	UK	Yes	Steering committee chair and meeting facilitator
Emergency Medicine[83]	2017	UK	Yes	Collaborator
Fibromyalgia[84]	2017	Canada	Yes	advised by JLA, JLA observer
Head and Neck Cancers[85]	2017	Canada - Alberta	Yes	Coach and Consultant
Hypertension[86]	2017	Canada	Yes	Advisor and facilitator
Miscarriage[87]	2017	UK	Yes	Steering committee chair and meeting facilitator
Pediatric Preventive Care[88]	2017	Canada	No	None
Tobacco Control/ Smoking Cessation[89]	2017	International	No	None
ADHD[90]	2018	Sweden	No	none
Alopecia - excl alopecia areata[44]	2018	UK	Yes	Co-chair and oversight
Aphasia following stroke[91]	2018	UK	yes	Conducted by JLA
Cystic Fibrosis[92]	2018	International	Yes	Facilitators
Dementia[93]	2018	Canada	Unclear	Unclear
Digital Tech in Mental Health[94]	2018	UK	Yes	Steering committee chair and meeting facilitator
Fragility Fractures[95]	2018	UK	Yes	Advisor
Lichen Sclerosus[96]	2018	UK (global reach)	Yes	Advisor
Lymphedema[97]	2018	UK	No	None
Pessary Use[98]	2018	UK	Yes	Conducted by JLA

Primary Care[99]	2018	UK	Yes	Facilitators
Type 2 DM[100]	2018	UK	Yes	Advisor and facilitator
Adult Malnutrition[101]	2019	UK	Yes	Advisor and facilitator
Anesthesia - Canada[102]	2019	Canada	Yes	Advisor
Blood Donation and Transfusion[103]	2019	UK	Yes	Guidance
Broken Bones of the Upper Limb[104]	2019	UK	Yes	Advisor and facilitator
Childhood learning difficulties[105]	2019	UK - Scotland	Yes	Steering committee chair and facilitator to help engagement
Frailty[106]	2019	Canada	No	None
Healthcare-associated Infections[107]	2019	UK	Yes	Facilitators
Human Milk Banking[108]	2019	Australia	No	None
Hyperacusis[109]	2019	UK (global reach)	Yes	Facilitator
Idiopathic Intracranial Hypertension[110]	2019	UK	Unclear	Unclear
Young People with Cancer[111]	2019	UK	Yes	Steering committee chair and meeting facilitator
Advanced Heart Failure[112]	2020	UK	Yes	Advisor
Anorexia Nervosa[28]	2020	Canada	Yes	Guidance
Cardiac Surgery[113]	2020	UK (International partcipation)	Yes	Steering committee chair and administrative support
Health Service Delivery for Psoriasis[114]	2020	UK	Yes	Steering committee chair
Liver Glycogen Storage Disease[115]	2020	International	Yes	Advisor and facilitator
Pregnancy Hypertension[116]	2020	UK	Yes	Steering committee chair and meeting facilitator
Problematic Knee Arthroplasty[117]	2020	UK	Yes	Advisor and facilitator
Rare Musculoskeletal Diseases[118]	2020	UK	Yes	Advisor, facilitator and project manager
Retinoblastoma[119]	2020	Canada	No	None

Table 2: Barriers and Facilitators related to Stakeholder Engagement

Topics	Codes	Articles Reporting Weakness (N)	Articles Reporting Strength (N)	Articles Reporting as Mixed (N)
Factors Specific to Subject of PSP				
Isolation (n=9)	Isolated population - medical cause	7	0	0
	Isolated population - Stigma	2	0	0
Rarity (n=1)	Rare disease	0	0	1
	Rare expertise	1	0	0
Disease Burden (n=10)	Spectrum of disease representation	6	0	0
	Difficulty engaging caregivers	4	0	0
Patient Group Definition(n=7)	Lack of belonging to well defined group	5	0	0
	Difficulty defining diagnosis/population	2	0	0
Factors Related to Social Condition of Affected Population				
Diversity (n=40)	Ethnic minority representation	20	2	0
	Low SES /Education representation	7	0	0
	Geographic representation	6	6	2
	Sex/Gender/LGBTQ representation	8	1	0
	Age representation	11	3	0
	Targeting population already accessing care/ advocacy	6	0	0
Factors Related to Health Care Providers				
HCP Involvement (n=8)	Community allied heath professional involvement	2	1	0
	Community general practitioner involvement	3	1	0
	Health care provider involvement	3	0	0

Factors Related to Civil Society Context				
Advocacy (n=9)	Presence of Strong Advocacy group	0	2	1
	Lack of Advocacy group	4	0	0
	Lack of advocacy group participation	2	0	0
Academic Research Culture		1	0	0
Factors Related to Mechanism of Engagement				
Steering Committee Form and Function (n=20)	Effective Introductory meeting	0	1	0
	Broad stakeholder involvement	1	10	1
	Collaboration between groups	0	3	0
	Autonomy	0	3	0
	Web based meeting technology	0	1	0
Advertising (n=9)	Social media use	2	2	0
	Advertising in traditional channels	0	2	0
	Use of specific targeting of hard to reach group	0	4	0
Survey Administration (n=23)	Multimodal communication methods	0	4	0
	Use of online survey	8	3	2
	Face to face meetings for uncertainty gathering	0	4	0
	Direct HCP involvement in survey distribution	0	2	0
	Use of incentives	0	1	1
	Encouraged group responses	0	1	0

Table 3: Barriers and Facilitators Related to Uncertainty Gathering

Topics	Code	Articles Reporting Weakness (N)	Articles Reporting Strength (N)
Survey Uptake (n=12)	Inconsistent collection methods	1	0
	Survey response	4	5
	Large proportion of blank surveys	2	0
Role of Research (n=10)	Difficulty with understanding concept of uncertainty	3	0
	Lack of understanding around what research can achieve	2	0
	Challenges with question formulation	8	0
	Safety concerns with limited means to follow-up	1	0
	Facilitator to answer mail/reframe complaints as questions	0	3
Scope (n=6)	Degree of concordance in scope of stakeholder questions	1	1
	Over-representation of researcher perspective	1	0
	Inappropriate scope of questions	3	0
Source of Uncertainties (n=2)	Recycled uncertainties from another PSP	1	0
	Flexibility with adding uncertainty at workshop	0	1
	Questions answered by research included anyway	1	0

Table 4: Barriers and Facilitators Related to Collation and Categorization

Topics	Codes	Articles Reporting Weakness (N)	Articles Reporting Strength (N)
Data Handling (n=6)	Large number of questions submitted	2	0
	Resource intensive	2	0
	Difficulty with categorization	4	0
Question Formulation (n=15)	Tension between broad and specific questions	5	0
	Retention of granular question detail	0	3
	Collating specific questions into broad ones	2	3
	Converting submissions into research questions	2	0
	Using specific format to formulate questions	0	1
Literature Assessment (n=7)	Use of "indicative uncertainties"	0	3
	Limited analysis	2	0
	Difficulty assessing literature	1	0
	Adapting concept of "treatment uncertainty" to broader scope	1	0
	Use of expert information specialist	0	1

Table 5: Barriers and Facilitators Related to Interim Prioritization

Topics	Codes	Articles Reporting Weakness (N)	Articles Reporting Strength (N)
Prioritization Tools (n=2)	Difficulty with concept of prioritization	1	0
	Use of simplified tool for prioritization	0	1
	Creation of education tool	0	1
	Difficulty developing appropriate tool for prioritization	1	0
Fairness/Equity (n=10)	Broader questions assigned higher priorities	4	0
	Actively tried to achieve fair/ equitable list of priorities	0	3
	Disparity in interim priorities between stakeholder groups	2	0
	More frequent questions prioritized	1	0

Table 6: Barriers and Facilitators and Related to Final Prioritization

Topics	Codes	Articles Reporting Weakness (N)	Articles Reporting Strength (N)
Attendance (n=14)	Poor attendance	4	0
	Representativeness of attendees	4	4
	Adapting workshop design to enhance participation	0	4
Discussion (n=22)	Availability of contextual information	1	1
	Quality of discussion	5	9
	Balance between large plenary sessions and small group discussions	0	1
	Use of facilitators	0	7
Record keeping (n=1)	Poor record keeping of workshop discussion	1	0
	Poor record keeping of voting/participation	1	0
Final Prioritization (n=12)	Move away from patient -centred questions	1	0
	Allowed beyond scope questions	1	1
	Only broad questions prioritized in top 10	3	3
	Consensus	2	3

Chapter 3

Research Priorities in Myelodysplastic Syndromes: A Survey of Patients, Caregivers and Clinicians

This manuscript is formatted for the American Journal of Hematology (not submitted)

Authors:

Grace Christou MD, MSc, FRCPC; Faculty of Medicine, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON, Canada

Ian D Graham PhD, FCAHS, FNYAM, FRSC; School of Epidemiology and Public Health, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON

Deepro Chowdhury MD; Faculty of Medicine, University of Ottawa

Dean Fergusson PhD; School of Epidemiology and Public Health, University of Ottawa, The Ottawa Hospital Research Institute, Ottawa, ON, Canada

Justin Presseau PhD; School of Epidemiology and Public Health, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON

Mitchell Sabloff MD, MSc, FRCPC; Faculty of Medicine, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON, Canada

Rena Buckstein MD, FRCPC; Division of Medical Oncology and Hematology, Odette Cancer Centre, Sunnybrook Health Sciences Centre, Toronto, ON, Canada

Cindy Anthony, Aplastic Anemia and Myelodysplasia Association of Canada, King City, ON, Canada

Gwen Barry, Aplastic Anemia and Myelodysplasia Association of Canada, King City, ON, Canada

Natasha Kekre MD, MPH, FRCPC; Faculty of Medicine, University of Ottawa, Ottawa Hospital Research Institute, Ottawa, ON, Canada

Statement of Contributions:

I designed this study in its entirety with feedback and guidance from supervisors and co-authors. Dr. Deepro Chowdhury contributed to coding verification.

Introduction:

Myelodysplastic syndromes (MDS) are a group of heterogeneous bone marrow malignancies characterized by morphologic and functional abnormalities in blood cell production and low blood counts[9] leading to reduced functional capacity, recurrent serious infections and bleeding. The presentation of MDS ranges from isolated mild to severe, but in all cases, a diagnosis of MDS carries a high risk for progression to acute myeloid leukemia (AML), a diagnosis that carries high morbidity and mortality.

An estimated 40,000 Canadians are living with MDS, and almost 4,000 new cases are diagnosed annually[120]. MDS has a median age of diagnosis of 70 years and an incidence that rises sharply with age. The incidence among those over 60 is 5 to 6 times that of the general population[9,11]. MDS is curable only by allogeneic stem cell transplantation[12], a highly toxic treatment for which only a minority of patients are eligible due to age and comorbidities. Thus, MDS treatment focuses on improving quality of life and slowing the progression towards acute leukemia.

The Need for Research Priority Setting in MDS:

While clinical care for MDS is largely focused on symptom management, the research landscape does not reflect this emphasis. The genetic basis of MDS is an area of intense research[17] and although the last decade has seen improvements in our understanding of disease biology, advances have not yet resulted in a fundamental shift in the treatment and management paradigm of MDS. At the same time, many aspects of MDS care that could enhance quality of life, such as transfusion optimization and iron chelation, have not been adequately studied and are extrapolated from other areas of hematology[19]. Other areas such as healthcare delivery, end of life care[20] and shared decision-making tools also remain essentially untapped[18]. Importantly, amid calls to focus research on the patient (i.e. patient oriented research) in choosing meaningful research outcomes, the focus in most studies in hematologic oncology continues to be on end-points that are of little or no value to patients[21].

Given these factors, engaging patients, their caregivers and clinicians in a priority setting partnership to identify the areas of greatest research need is both a worthwhile and timely endeavour in MDS. The James

Lind Alliance (JLA) approach to research priority setting follows a systematic and rigorous approach[22]. Established in 2004 with the goal of ensuring that health researchers and funders are aware of the issues that are most important to patients and clinicians, the JLA approach engages patients, their caregivers and treating clinicians to identify and prioritize research questions in a scientifically sound and transparent process. To date, it has been used by over 100 groups[32]. The Canadian MDS Priority Setting Partnership was established in December 2017 with the aim of identifying research questions that can shape national priorities in MDS research based on the JLA method. Here we report the results of the initial survey of this process and our experience conducting it.

Methods:

The Canadian MDS Priority Setting Partnership included hematologists mainly from Ontario in partnership with the Aplastic Anemia and Myelodysplasia Association of Canada (AAMAC). The 7-member steering committee included 4 hematologists and 3 members of AAMAC, one of whom was a patient with a lived experience of MDS.

Survey Design:

Separate surveys were designed for clinicians and non-clinicians to collect demographic and disease-specific information (Appendix 1). General demographic information questions with drop down menus were presented first, followed by open-ended questions to elicit MDS research questions and then disease specific questions with drop down menus. This was done to ensure that we had some context for our results and to assess our ability to reach diverse populations. Placing simple questions at the start of the survey also provided respondents a chance to familiarize themselves with the platform and format before being presented with more sophisticated questions about research which require a deeper level of thought and engagement. We opted to defer disease specific questions until later in the survey so as not to overburden respondents who were only interested in having their thoughts and questions heard. Questions specifically aimed at gathering MDS research questions were divided into 5 domains (diagnosis, prognosis, symptoms,

effects of treatment and healthcare delivery) and provided examples of items that fall into each category. This was done as focused questions have been shown to elicit more responses and more usable information than very broad, open ended questions[121]. Of note, none of the questions were mandatory and respondents were encouraged to submit their answers where they felt most appropriate. The surveys were piloted on 5 people with lived experience of MDS and 5 clinicians (nurses, pharmacists and hematologists) including French speakers before being finalized and administered. Translation to French was undertaken by a certified translator.

Survey Population Sample:

The survey was open to any adult over 18 years of age living in any Canadian province or territory and self-identifying as a person living with MDS, a friend or family member of someone living with MDS (i.e. person with lived experience of MDS) or either a pharmacist, nurse or physician involved in providing care to people living with MDS. This was an open survey; thus not possible to calculate an associated sample size or response rate.

Survey Administration:

Survey administration was adapted from Dillman's Total Design Method[121]. Surveys were administered online via Lime Survey™ in both English and French and could be accessed via the bilingual Canadian MDS PSP website. Our goal was to make the surveys accessible across Canada. The website was specifically designed to introduce the research team, explain the scope of work and explain how survey data would be used. The survey was open between September and December 2018. Both the parent website and survey were hosted by The Ottawa Hospital Research Institute (OHRI) and all data were collected and housed on OHRI servers in accordance with federal, provincial and institutional privacy laws and regulations. Paper copies of the survey were available upon request, although we did not receive any requests for them. The survey had an open format with no limit to the number of submissions by each respondent.

Advertising:

The survey was advertised through AAMAC and its partner organizations, the Canadian Hematology Society, Canadian Blood and Marrow Transplant Group (now Cell Therapy Transplant Canada), the Canadian Association of Nurses in Oncology and the Canadian Association of Pharmacy in Oncology through direct emails and through organizations' social media channels. Bilingual posters were displayed in malignant hematology, general hematology and thrombosis clinics at The Ottawa Hospital. AAMAC had access to 209 contacts on its regular mailing list at the time. Paid advertising in English and French on Facebook® and Google Ads® was also purchased to expand the reach of the PSP. For Facebook, this was done through promoted posts from The Ottawa Hospital account and was targeted to adults living in Canada, who have liked/followed health or science/research related pages. Google Ads® were targeted to people searching for relevant terms including MDS, myelodysplastic syndrome, dysplasia, pre-leukemia, anemia from anywhere within Canada. Advertisements ran on each platform for 2-3 weeks until the funds allocated for advertising were exhausted.

Data processing:

The raw qualitative data were extracted and coded using QSR International Nvivo™ (version 12) by one investigator (GC) and coding was verified by a second investigator (DC). For responses submitted as anecdotes or complaints, we coded these as long as there was an identifiable focus that could be turned into a research question. Codes were then grouped into subdomains and themed into domains to facilitate interpretation and recorded in Microsoft Excel®. Frequency distributions were used to describe the data.

Ethics Approval:

This study was approved by The Ottawa Health Science Network Research Ethics Board.

Reporting of Results:

Results of the survey are reported using the CHERRIES reporting checklist for surveys[122].

Results:

Survey respondents: Overall, 163 surveys were at least partially completed (Tables 1 and 2). Of these, 49 were from clinicians and 114 from patients and caregivers (69 patients and 41 caregivers). Ten surveys were submitted in French. After excluding surveys submitted without any research questions, 63 surveys were suitable for analysis, resulting in 203 codable research questions about MDS. Respondent characteristics are shown in Tables 1 and 2 for people with a lived experience of MDS and clinicians respectively. Surveys were received from every province (except PEI). There were no responses received from any of the territories. Residents of Ontario and Quebec accounted for 47.8% of all surveys submitted (57% of people with lived experience and 26.5% of clinicians) and 71.4% of all surveys that included at least one MDS research question. Women comprised the majority of respondents to each survey. Respondents who submitted at least one research question were mostly people living with MDS (66.7%), in large and medium sized urban communities (83.3%) in Ontario (62.5%). Among clinicians, the group with at least one research question was overwhelmingly comprised of hematologists (93%), mostly practicing in academic centres in large or medium urban areas in central Canada.

Survey Results: Collated qualitative data (research questions posed) were then coded and categorized, resulting in 203 codable questions about 57 specific subdomains which were then grouped into 14 domains. Managing symptoms of MDS was the domain into which the highest number of codes fell. Thirty-seven questions accounting for 18% of all submissions were in this domain, 30 of them from people with a lived experience of MDS and 7 from clinicians. Symptoms of MDS comprised 10 different subdomains (Table 3), fatigue being the most commonly asked about (17 submissions total). Living with MDS (15.8%), communication (11.3%) and questions about diagnosis (9.9%) were the second, third and fourth most common domains into which individual queries segregated.

Living with MDS included 11 different subcategories ranging from the role of diet and exercise to dental care, sexuality and end of life. Four clinician questions fell into this category (2 on enhancing quality of

life and 2 on end of life), whereas people with lived experience submitted 28 questions about diet, improving quality of life, exercise, prevention of infections, dental and sexual health.

Communication was the third most commonly asked about domain, and the most common subdomain, comprising 11.3% of all submissions, 13% of questions from people living with MDS and 7.7% of questions from clinicians.

Questions surrounding diagnosis of MDS were the fourth most common domain of questions (9.9%). People with lived experience were exclusively interested in possibilities for earlier diagnosis and genetic factors (5.8% of questions) whereas questions from clinicians also touched upon understanding of MDS variants, differentiating between MDS and other bone marrow failure disorders and whether morphologic bone marrow characterization was still relevant today in the diagnosis of MDS (9.9% of questions).

Access to care was the fifth most common domain, accounting for 9.3% of total submissions (10% among patients and caregivers and 7.7% among clinicians). People living with MDS and their loved ones submitted questions about access to MDS specialists, a national strategy for treating MDS and better integration among the many healthcare providers involved in patients' care. Better healthcare integration was the main question submitted by clinicians within this broader domain (4 of 5 questions total).

Process Challenges: Our work on developing and administering a national survey to elicit patient and clinician input on research priorities is novel in the field of hematologic malignancy. Along the way, we encountered a number of challenges worth reporting to help inform the work of other groups. First, our partner AAMAC was in the midst of a website rebuild, thus making it difficult for our survey to be hosted on their server. This necessitated the building of a new bilingual website to introduce the PSP and host the survey. Second, this type of work where patients are equal participants in the work is relatively novel and thus, there are no specific mechanisms within existing research ethics and privacy frameworks to streamline requirements for those embarking on such endeavours. This necessitated a number of consultations with privacy officers, institutional ethics boards, IT and communications experts to plan each aspect of the

survey and the advertising around it. Still, a number of unexpected hurdles emerged. For example, our partner organization had direct contact information of only about 200 patients, making it difficult to reach a wide audience directly. AAMAC kindly disseminated the survey via other, larger partner organizations based in the United States, but it is unclear how many Canadians with MDS these reached. This necessitated that we purchase paid advertising, which we had not initially anticipated. During that process, we had to re-design our initial advertising to comply with new Facebook advertising policies introduced around the same time as our launch. Facebook advertising policies prohibit content that asserts or implies personal attributes, including those about a person's medical condition[123]. Furthermore, while we were able to advertise and discuss the ongoing survey with patients visiting hematology clinics at our institution, we were unable to expand this to include major MDS treatment sites elsewhere because each institution would need to seek approval from its own REB, thus significantly increasing the time and personnel resources needed.

In terms of survey responses, we received many surveys where demographic information was completed, but no research questions were submitted. Others offered anecdotes detailing personal experiences with MDS and medical care for MDS. A concerted effort was made to honour participants' stories and struggles by coding them as long as there was a specific issue that could possibly be addressed by research.

Discussion:

Our PSP has successfully administered a national survey to people affected by MDS, their loved ones and clinicians caring for them. Forty-eight people with a lived experience of the condition and 15 clinicians responded generating 203 MDS questions that can potentially be answered by research. To our knowledge, this is the first survey of research priorities simultaneously targeting clinicians and patients in the area of hematologic malignancy. The questions submitted by survey participants covered a wide array of areas relevant to MDS spanning from diagnosis to care delivery. Because JLA PSPs often involve an open call

for research question submissions, it is uncertain how many people see these calls on various media and interact with them. This means that the denominator for calculating a response rate cannot be known. Despite that, when compared with other Canadian-based priority setting partnerships, our absolute respondent numbers were low. Other Canadian PSPs have had numbers ranging from 75 for gestational diabetes[124] to almost 1217 for dementia[93], with most in the 200-300 range[28,68,86,125]. When taking into account the relative rarity of MDS compared to other conditions, however, the number of responses, particularly from those living with MDS and their caregivers, compare favourably. For example, the Canadian hypertension PSP[86] received 386 responses for a condition that affects roughly 7.7 million Canadians[126], compared to an estimated 40,000 Canadians affected by MDS[120]. The large proportion of surveys submitted without any research questions is also not unusual within the sphere of similar studies and is reported by several PSPs including head and neck cancers in Alberta[125] and acne in the United Kingdom[71].

As expected, there were differences in the research priorities expressed by clinician and non-clinician respondents. People with a lived experience with MDS asked more questions about symptoms of MDS and how to live better with MDS than clinicians, whereas clinicians asked about diagnosis and more accurate prognostic tools, both areas of very intensive research activity. Both clinicians and people living with MDS were uniformly interested in improving communication around this complex and progressive condition. This confirms previous findings that a high proportion of patients did not fully understand the nature of their diagnosis or goals of treatment and that this was a source of immense distress[13,127]. In fact, many of the anecdotal submissions we received centred around the mental anguish caused by communication challenges. The strong focus on managing symptoms reflects the fact that MDS is experienced as a chronic condition, not imminently fatal in most cases, but significantly impairing the quality of life of people living with it.

Our study had a number of strengths and limitations which are common with survey studies. In terms of representativeness, patients with lower risk MDS represent approximately two thirds of all those living with

MDS, so it is not surprising that those with lower risk MDS comprised the largest group of respondents. Additionally, people with higher risk MDS often suffer profound fatigue, spend many hours a week receiving blood tests, blood transfusions, hypomethylating agents and being treated for, or recovering from complications of treatment. Friends and family are often recruited to help with care, leaving less time and energy available to both those diagnosed with MDS and their caregivers to participate in a survey that requires a significant level of intellectual engagement. Reflecting on other PSPs that targeted frail, medically complex populations[43,45,93], in-person interviews in places where people are receiving care, encouraging group submissions and generous coding of anecdotes were all successfully used to enhance participation. In terms of inclusiveness, although the question we used to assess gender identity was approved by the REB, in hindsight, it should have been formulated using more inclusive lamguage. Geographically, submitted research questions came mostly from urban centres in Ontario and Quebec although more respondents from other provinces submitted partially completed surveys. We had low participation from provinces and territories with a high proportion of sparsely populated rural communities. This reflects the challenges posed by the vast Canadian landscape which significantly limits equal access to many services. For example, high speed internet access is unavailable or prohibitively expensive in many rural areas[128]. Additionally, Canadians living in remote areas have lower access to primary and subspecialty medical care[129]. Beyond geographic barriers, a study of rural and remote communities of northern British Columbia suggests that rural residents may have a strong preference for participating in research projects that have a local impact[130]. In our study, rural respondents comprised almost 9% of those who submitted a partially completed survey, but none contained any research questions. Engaging with rural populations requires specific outreach that was beyond the scope of our current work, but is important in improving MDS care on a national and provincial/territorial level.

On the clinician side, aside from one submission by a nurse, only hematologists participated in our survey, despite the fact that MDS patients tend to be elderly and highly co-morbid, necessitating the care of several practitioners in a variety of settings[131–133]. Further highlighting this, access to care, including improved

access to MDS specialists, establishing a national standard for MDS care and better integration between healthcare providers was the fifth most common domain in our study. Despite these challenges, our study had a number of strengths. We followed best practices in survey development with a dedicated website portal and partnerships with recognized partner organizations, enhancing trust in our initiative. Surveys were piloted with the help of people living with MDS, physicians, nurses and pharmacists. We used a rigorous advertising strategy for a rare disease and analyzed data using an inclusive coding strategy. Our results show that we engaged with a diversity of views. Research questions submitted covered almost every aspect of MDS care, including issues that would be pertinent to rural populations.

Our survey identified that treating symptoms and improving communication are top research priorities for people living with MDS. Overall, our work represents an important first step towards defining the MDS research agenda in Canada with strong input from people whose lives have been touched by this diagnosis. We identified limitations to engagement at this stage will shape our strategy for the interim and final prioritization stages, such that we can capture a wider range of perspectives.

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Tables and Figures:

Table 1: Respondent Characteristics of People with lived experience of MDS Survey

		Total Respondents *N=114*	**Respondents with ≥ 1 research question** *N= 48*
Respondent Role *n (%)*	Person Living with MDS	69 (60.5)	32 (66.7)
	Friend/ Family Member	41 (36.0)	16 (33.3)
	No Answer	4 (3.5)	0 (0.0)
Survey Language *n (%)*	English	108 (94.7)	45 (93.8)
	French	6 (5.3)	3 (6.3)
Age	Mean yrs (range)	64 years (18-84)	67 years (41-82)
Gender *n (%)*	Man	36 (31.6)	17 (35.4)
	Woman	75 (65.8)	31 (64.6)
	Other	0 (0.0)	0 (0.0)
	No Answer	3 (2.6)	0 (0.0)
Province of Residence *n (%)*	Newfoundland and Labrador	1 (0.9)	0 (0.0)
	Prince Edward Island	0 (0.0)	0 (0.0)
	Nova Scotia	7 (6.1)	3 (6.3)
	New Brusnwick	5 (4.4)	0 (0.0)
	Quebec	13 (11.4)	4 (8.3)
	Ontario	52 (45.6)	30 (62.5)
	Manitoba	2 (1.8)	0 (0.0)
	Saskatchewan	2 (1.8)	1 (2.1)
	Alberta	10 (8.8)	4 (8.3)
	British Columbia	16 (14.0)	6 (12.5)

	No Answer	6 (5.3)	0 (0.0)
Community Size *n(%)*	Large Urban	48 (42.1)	25 (52.1)
	Medium Urban	31 (27.2)	15 (31.3)
	Small Urban	17 (14.9)	5 (10.4)
	Rural	10 (8.8)	3 (6.3)
	No Answer	8 (7.0)	0 (0.0)
IPSS Score *n (%)*	Low	18 (15.8)	15 (31.3)
	Intermediate - 1	7 (6.1)	7 (14.6)
	Intermediate - 2	6 (5.3)	5 (10.4)
	High	7 (6.1)	7 (14.6)
	Other	2 (1.8)	2 (4.2)
	Don't know	10 (8.8)	9 (18.8)
	No Answer	64 (56.1)	3 (6.3)
Type of Setting where MDS Care is Received *n (%)*	Academic Centre	26 (22.8)	23 (47.9)
	Community Practice	16 (14.0)	13 (27.1)
	Don't know	10 (8.8)	8 (16.7)
	No Answer	62 (54.4)	4 (8.3)

Table 2: Respondent Characteristics of Clinician Survey

		Total Respondents *N=49*	**Respondents with ≥ 1 research question** *N= 15*
Respondent Role *n (%)*	Hematologist	31 (63.3)	14 (93.3)
	Other physician	1 (2.0)	0 (0.0)
	Nurse	8 (16.3)	1 (6.7)
	Pharmacist	5 (10.2)	0 (0.0)
	Other	4 (8.2)	0 (0.0)
	No Answer	0 (0.0)	0 (0.0)
Survey Language *n (%)*	English	45 (91.8)	13 (86.7)
	French	4 (8.2)	2 (13.3)
Time In Practice	Mean yrs (range)	8.6 yrs (1-25)	9.7 (2-25)
Gender *n (%)*	Man	17 (34.7)	6 (40.0)
	Woman	31 (63.3)	9 (60.0)
	Other	1 (2.0)	0 (0.0)
	No Answer	0 (0.0)	0 (0.0)
Province of Residence *n (%)*	Newfoundland and Labrador	0 (0.0)	0 (0.0)
	Prince Edward Island	0 (0.0)	0 (0.0)
	Nova Scotia	0 (0.0)	0 (0.0)
	New Brunswick	0 (0.0)	0 (0.0)
	Quebec	3 (6.1)	3 (20.0)
	Ontario	10 (20.4)	8 (53.3)
	Manitoba	0 (0.0)	0 (0.0)
	Saskatchewan	0 (0.0)	0 (0.0)
	Alberta	1 (2.0)	1 (6.7)

	British Columbia	0 (0.0)	0 (0.0)
	No Answer	35 (71.4)	3 (20.0)
Practice Community Size *n (%)*	Large Urban	12 (24.5)	10 (66.7)
	Medium Urban	2 (4.1)	2 (13.3)
	Small Urban	0 (0.0)	0 (0.0)
	Rural	0 (0.0)	0 (0.0)
	No Answer	35 (71.4)	3 (20.0)
Practice Setting *n (%)*	Academic Centre	10 (20.4)	9 (60.0)
	Community Hospital Practice	4 (8.2)	3 (20.0)
	No Answer	35 (71.4)	3 (20.0)

Table 3: Research Question Response Frequencies by Domain and Subdomain

		Lived Experience N= 138		***Clinicians*** N= 65		***Total*** N=203	
Domain	**Subdomain**	**Subdomain**	**Domain**	**Subdomain**	**Domain**	**Subdomain**	**Domain**
Stem Cell Transplant	*Optimal AlloHCT*	1 (0.7)	1 (0.7)	0 (0)	0 (0)	1 (0.5)	1 (0.5)
Social Burden	*Caregiver Support*	1 (0.7)	1 (0.7)	0 (0)	1 (1.5)	1 (0.5)	2 (1.0)
	Impact on Family	0 (0)		1 (1.5)		1 (0.5)	
Transfusion	*Alloimmunization*	1 (0.7)	3 (2.2)	0 (0)	3 (4.6)	1 (0.5)	6 (3.0)
	Transfusions	1 (0.7)		3 (4.6)			
	Iron Chelation	1 (0.7)		0 (0)		1 (0.5)	
Treatment Evaluation	*Cost Effectiveness*	0 (0)	2 (1.4)	1 (1.5)	5 (7.7)	1 (0.5)	7 (3.4)
	Drugs	1 (0.7)		0 (0)		1 (0.5)	
	Effect of NGS on treatment	0 (0)		1 (1.5)		1 (0.5)	
	Targeted Therapy	0 (0)		2 (3.1)		2 (1)	
	Treatment Sequencing	1 (0.7)		1 (1.5)		2 (1)	

Symptoms	*Appetite*	5 (3.6)	30 (21.7)	0 (0)	7 (10.8)	5 (2.5)	37 (18.2)
	Bleeding	2 (1.4)		0 (0)		2 (1)	
	Depression	1 (0.7)		1 (1.5)		2 (1)	
	Effects of Anemia	2 (1.4)		0 (0)		2 (1)	
	Fatigue	13 (9.4)		4 (6.2)		17 (8.4)	
	Nausea	0 (0)		1 (1.5)		1 (0.5)	
	Neuropathy	2 (1.4)		0 (0)		2 (1)	
	Pain	1 (0.7)		0 (0)		1 (0.5)	
	Sweats	3 (2.2)		1 (1.5)		4 (2.0)	
	Weight Loss	1 (0.7)		0 (0)		1 (0.5)	
Access to Care	*HCP Integration*	3 (2.2)	14 (10.1)	4 (6.2)	5 (7.7)	7 (3.4)	19 (9.4)
	MDS Specialist Access	6 (4.3)		1 (1.5)		7 (3.4)	
	National Strategy	4 (2.9)		0 (0)		4 (2.0)	
	Role of Electronic Health Tracking	1 (0.7)		0 (0)		1 (0.5)	
Communication	*Communication*	18 (13)	18 (13.0)	5 (7.7)	5 (7.7)	23 (11.3)	23 (11.3)
Alternative Therapies	*Cannabis*	1 (0.7)	6 (4.3)	0 (0)	2 (3.1)	0 (0)	8 (3.9)
	Holistic Drugs	5 (3.6)		1 (1.5)		6 (3.0)	

	Complementary Treatments	0 (0)		1 (1.5)		1 (0.5)	
Living with MDS	*Dental Issues*	1 (0.7)	28 (20.3)	0 (0)	4 (20.3)	1 (0.5)	32 (15.8)
	Diet	9 (6.5)		0 (0)		9 (4.4)	
	EOL Care	0 (0)		2 (3.1)		2 (1)	
	Enhance QoL	6 (4.3)		2 (3.1)		8 (3.9)	
	Exercise	5 (3.6)		0 (0)		5 (2.5)	
	Group Support	2 (1.4)		0 (0)		2 (1)	
	Measuring QoL	1 (0.7)		0 (0)		1 (0.5)	
	Preventing Infection	2 (1.4)		0 (0)		2 (1)	
	Immunizations	1 (0.7)		0 (0)		1 (0.5)	
	Sexuality	1 (0.7)		0 (0)		1 (0.5)	
Cytopenias	*Increasing Platelets*	1 (0.7)	7 (5.1)	0 (0)	3 (5.1)	1 (0.5)	10 (4.9)
	Effect of Anemia	1 (0.7)		0 (0)		1 (0.5)	
	Relationship to Symptoms	2 (1.4)		2 (3.1)		4 (2.0)	
	Treatment of Anemia	3 (2.2)		1 (1.5)		4 (2.0)	

Diagnosis	*Early Diagnosis*	4 (2.9)	8 (5.8)	3 (4.6)	12 (18.5)	7 (3.4)	20 (9.9)
	Better Understanding of Variants	0 (0)		1 (1.5)		1 (0.5)	
	Differentiating Hypocellular MDS from Marrow Failure	0 (0)		1 (1.5)		1 (0.5)	
	Genetic Factors	4 (2.9)		6 (9.2)		10 (4.9)	
	Role of Morphology	0 (0.0)		1 (1.5)		1 (0.5)	
Hypomethylating Agents	*Coaching Through HMA*	0 (0.0)	3 (2.2)	1 (1.5)	8 (12.3)	1 (0.5)	11 (5.4)
	Home HMA	0 (0.0)		1 (1.5)		1 (0.5)	
	Pain with HMA (Azacitidine)	1 (0.7)		1 (1.5)		2 (1)	
	Post HMA Failure	1 (0.7)		2 (3.1)		3 (1.5)	
	Predictors of HMA Response	1 (0.7)		3 (4.6)		4 (2.0)	
Pathobiology	*Causes*	9 (6.5)	10 (7.2)	0 (0)	2 (3.1)	9 (4.4)	12 (5.9)

	Role of Inflammatory Cytokines	0 (0.0)		1 (1.5)		1 (0.5)	
	Relationship with Comorbidities	1 (0.7)		1 (1.5)		2 (1)	
Prognostics	*More Accurate Prognostic Score*	7 (5.1)	7 (5.1)	8 (12.3)	8 (12.3)	15 (7.4)	15 (7.4)

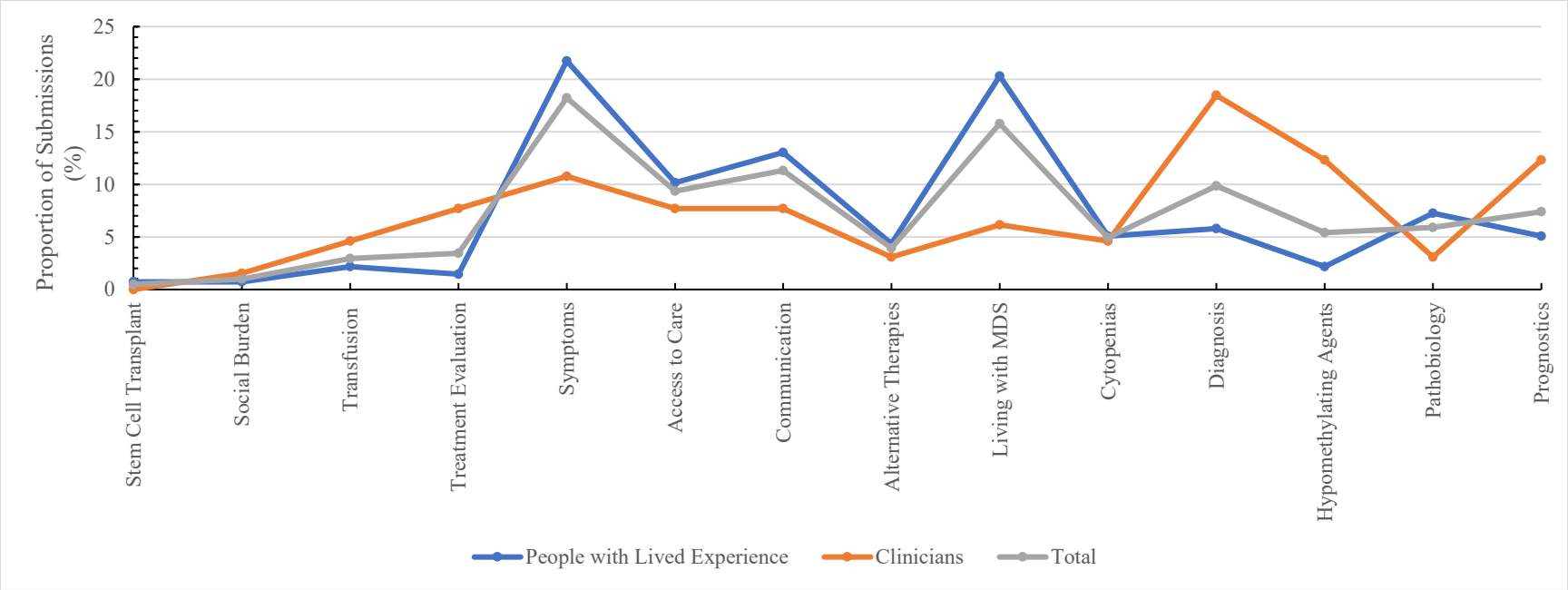

Figure 1: Distribution of Research Questions by Doma

APPENDIX 1A

MDS Patients and Caregivers Survey

Introduction:

Thank you for your interest in the Canadian Myelodysplastic Syndromes Priority Setting Partnership Survey. This project is supported by the Ottawa Hospital Research Institute, the Strategy for Patient Oriented Research and the Ottawa Blood Disease Centre in Partnership with the Aplastic Anemia and Myelodysplasia Association of Canada.

We are a group of researchers, physicians, patients and advocates who are interested in hearing from people who have experience with myelodysplastic syndromes (MDS).

We are asking people with MDS, their family members, partners, friends, caregivers and healthcare professionals to answer 5 questions about what they think are important questions for future research in MDS.

We want to know what you think are questions that should be answered by research. You do not have to check whether your questions have already been researched, we will do that at a later stage. If answers to different questions seem to overlap, record them wherever you feel most appropriate. Once you have started the survey, you may save your answers and return to it as many times as you would like until you are ready to submit your answers.

Your responses are anonymous and will only be used for this project. Your information is stored on servers at The Ottawa Hospital and will never be shared or sold to third parties.

Part I Demographics:

Please help us get to know you better

1- Which of the following best describes you?
   a. Person living with MDS
   b. Friend or family member of a person living with MDS

2- Gender:
   a. Man
   b. Woman
   c. Other

3- What age (in years) were you at your last birthday?

Free type (numbers)

4- Which Canadian Province or Territory do you currently live in?

Drop down menu options:

Newfoundland and Labrador, Prince Edward Island, Nova Scotia, New Brunswick, Quebec, Ontario, Manitoba, Saskatchewan, British Columbia, Nunavut, Yukon, Northwest Territories

5- Which of the following best describes the community you currently live in?
   a. Large Urban Population Centre (Population size 100,000 or more)
   b. Medium Population Centre (Population size 30,000-99,999)
   c. Small Population Centre (Population size 1,000-29,999)
   d. Rural Area (Population size less than 1,000)

Part II- Questions about MDS Care

1- What questions about MDS ***diagnosis*** would you like to see answered by research?
[These may include questions about the tests needed for diagnosis, how the diagnosis is communicated to patients and their families or the experience of being diagnosed with MDS.]

2- What questions about MDS ***prognosis*** would you like to see answered by research?
(Prognosis is the course a particular condition is expected to take)
[These may include questions about how things like overall health affects prognosis or how a prognosis is communicated to patients and their families]

3- What questions about ***symptoms of MDS*** would you like to see answered by research?
[These may include questions about fatigue, lack of appetite or treatments that can be used to improve them]

4- What questions about the ***effects of treatment for MDS*** would you like to see answered by research?
[These may include questions about transfusion, chemotherapy, stem cell/bone marrow transplant or non-medicinal treatments like exercise, nutrition etc.]

5- What questions about ***MDS care delivery*** would you like to see answered by research?
[These may include questions about how healthcare teams work together, access to treatments and services in rural or urban settings etc.]

Part III- Diagnosis and Treatment Details
We would like to make sure we hear from a broad segment of people affected by MDS. The following questions will help us understand who we have been able to reach so far.

1- Approximately what year were you (or your loved one) diagnosed with MDS?
Free type (year)

2- What type of MDS do you (or your loved one) have?
   a. Low risk
   b. Intermediate risk-1
   c. Intermediate risk-2
   d. High Risk

e. Don't know
f. Other (Please specify)

3- Which of the following best describes the community you (or your loved one) currently receive medical care for MDS?
   a. Large Urban Population Centre (Population size 100,000 or more)
   b. Medium Population Centre (Population size 30,000-99,999)
   c. Small Population Centre (Population size 1,000-29,999)
   d. Rural Area (Population size less than 1,000)

4- Where do you (or your loved one) receive the majority of medical care for MDS?
   a. Academic Centre
   b. Community based practice
   c. I don't know

5- How far do you (or your loved one) have to travel to receive the majority of medical care for MDS?
   a. <1 hour
   b. 1-2 hours
   c. >2 hours

6- Which of the following treatments for MDS are you (or your loved one) currently receiving? Check all that apply
   a- Transfusions (red cells and/or platelets)
   b- Tranexamic acid (medication used to prevent/reduce bleeding)
   c- Eprex or Aranesp (medication used to stimulate red cell production)
   d- Iron chelator (medication that removes excess iron from the body)
   e- Azacitidine (also known as Vidaza)
   f- Lenalidomide (also known as Revlimid)
   g- Neupogen or Filgrastim or G-CSF (hormone that stimulates neutrophil or white cell production)
   h- Chemotherapy other than vidaza/azacitidine
   i- Stem cell/ bone marrow transplant
   j- Palliative or supportive treatment (treatment that helps with the symptoms of MDS)
   k- None
   l- I don't know
   m- Other (please specify)

7- Which of the following treatments for MDS have you (or your loved one) received in the past? Check all that apply
   a- Transfusions (red cells and/or platelets)
   b- Tranexamic acid (medication to prevent/reduce bleeding)
   c- Eprex or Aranesp (hormone that stimulates red cell production)
   d- Iron chelator (medication that removes excess iron from the body)
   e- Azacitidine (also known as Vidaza)
   f- Lenalidomide (also known as Revlimid)
   g- Neupogen or Filgrastim or G-CSF (hormone that stimulates neutrophil or white cell production)
   h- Chemotherapy other than vidaza/azacitidine
   i- Stem cell/ bone marrow transplant
   j- Palliative or supportive treatment (treatment that helps with the symptoms of MDS)
   k- None

l- I don’t know
m- Other (please specify)

Thank you for completing the survey.

For ongoing updates about the project please check in at [URL] regularly.

APPENDIX 1B

MDS Clinician Survey

Introduction:

Thank you for your interest in the Canadian Myelodysplastic Syndromes Priority Setting Partnership Survey. This project is supported by the Ottawa Hospital Research Institute, the Strategy for Patient Oriented Research and the Ottawa Blood Disease Centre in Partnership with the Aplastic Anemia and Myelodysplasia Association of Canada.

We are a group of researchers, physicians, patients and advocates who are interested in hearing from people who have experience with myelodysplastic syndromes (MDS).

We are asking people with MDS, their family members, partners, friends, caregivers and healthcare professionals to answer 5 questions about what they think are important questions for future research in MDS.

We want to know what you think are questions that should be answered by research. You do not have to check whether your questions have already been researched, we will do that at a later stage. If answers to different questions seem to overlap, record them wherever you feel most appropriate. Once you have started the survey, you may save your answers and return to it as many times as you would like until the survey closes on **(enter date)** or you are ready to submit your answers.

Your responses are anonymous and will only be used for this project. Your information is stored on servers at The Ottawa Hospital and will never be shared or sold to third parties.

Part I Demographics:

Please help us get to know you better

1- Which of the following best describes you?
   c. Hematologist
   d. Other physician
   e. Nurse
   f. Pharmacist
   g. Other health care professional (please specify)

2- Gender:
   a. Man
   b. Woman
   c. Other

Part II- Questions about MDS Care

6- What questions about MDS ***diagnosis*** would you like to see answered by research?

[These may include questions about the tests needed for diagnosis, how the diagnosis is communicated to patients or the experience of being diagnosed with MDS.]

7- What questions about MDS ***prognosis*** would you like to see answered by research?
[These may include questions about how things like overall health affects prognosis or how a prognosis is communicated to patients and their families]

8- What questions about ***symptoms of MDS*** would you like to see answered by research?
[These may include questions about fatigue, lack of appetite or treatments that can be used to improve them]

9- What questions about the ***effects of treatment for MDS*** would you like to see answered by research?
[These may include questions about transfusion, chemotherapy, stem cell/ bone marrow transplant or non-medicinal treatments like exercise, nutrition etc.]

10- What questions about ***MDS care delivery*** would you like to see answered by research?
[These may include questions about how healthcare teams work together, access to treatments and services in rural or urban settings etc.]

Part III- Practice Details

We would like to make sure we hear from a broad segment of clinicians who care for patient with MDS. The following questions will help us understand who we have been able to reach so far.

1- How long in years have you been caring for patients with MDS?
Free type (numbers)

2- Which Canadian Province or Territory do you currently practice in?
Drop down menu options:
Newfoundland and Labrador, Prince Edward Island, Nova Scotia, New Brunswick, Quebec, Ontario, Manitoba, Saskatchewan, British Columbia, Nunavut, Yukon, Northwest Territories

3- Which of the following best describes the community you currently practice in?
   a. Large Urban Population Centre (Population size 100,000 or more)
   b. Medium Population Centre (Population size 30,000-99,999)
   c. Small Population Centre (Population size 1,000-29,999)
   d. Rural Area (Population size less than 1,000)

4- What type of setting do you currently practice in?
   d. Academic centre
   e. Community hospital based practice
   f. Community clinic/ physician's office based practice

g. Other (please specify)

Thank you for completing the survey.

For ongoing updates about the project please check in at [URL] regularly.

Chapter 4

Integrated Discussion

My thesis focuses on the JLA process and is composed of two studies. The first study was a scoping review of the real world experience of using the JLA method for research priority setting, as reported by priority setting partnerships (PSPs) themselves. The second study involved conducting the first 3 stages of the Canadian MDS Priority Setting Partnership and describing our experience with the process and our preliminary results. Our goal in carrying out this work was to gain a deeper understanding of the implementation and methodological aspects of the JLA method in light of its growing popularity. Specifically, we looked at barriers and facilitators to using the JLA method and catalogued the ways in which various groups adapted the method. Our hope is that these studies taken collectively will provide guidance for other researchers and PSP groups as they embark on their own PSPs. Furthermore, we used some of our findings from the literature to guide implementation of our own JLA PSP in MDS, a complex hematologic malignancy requiring chronic multidisciplinary care.

In the first manuscript (Chapter 2) we analyzed our scoping review findings based on the stage of the JLA process. We then incorporated some of the findings from our review into our implementation of the JLA method, within the limits of available resources. The challenges we uncovered can be broadly classified into 3 areas: representation, data handling and usability of results. Here we review these themes and how our experience conducting our own JLA process (Chapter 3) relates to them.

Representation:

Representation is related to several factors which all relate to the difficulty engaging with equity deserving groups, whether the result of disability, race, ethnicity, linguistic, economic or geographic factors. Furthermore, poor demographic data collection made it difficult for PSPs to properly evaluate the diversity

of individuals, experiences and views that their priority setting exercise represented and thus, to contextualize their findings. In our own MDS PSP, we also faced a diversity challenge whereby despite reaching a number of pharmacists and nurses, none submitted their questions for MDS research. Likewise, most of the questions from patients and caregivers were submitted by urban dwellers with lower risk disease, receiving treatment at academic centers. We attempted to collect more detailed demographic and disease information so that we could properly contextualize our results and this worked well in helping us identify groups that were underrepresented, such that we can redouble our efforts to engage them in later stages of the process. Reaching groups which have been left out of traditional engagement approaches requires diversification of the uncertainty gathering phase of the JLA method beyond internet surveys and using alternative sampling methods such as purposive and snowball sampling[55].

There are no clear criteria for evaluating representativeness[48], but groups should carefully consider whether they have resources to reach certain special populations, how prevalent the condition of interest is among marginalized groups vs the "general population" and whether the differences between the priorities of these groups is likely to pose a threat to the validity of the results within the scope of the PSP[48]. Once they have done this, PSPs should set their own benchmarks for sample diversity, utilize methods known to be effective for specifically engaging their target groups, and evaluate their results against these targets.

One of the possible explanations for the observed persistent lack of diversity, is that advertising through social media and internet based surveys play a central role in collecting research questions for PSPs. While technology has facilitated the work of PSPs and can clearly enhance reach, it is not a panacea for engagement, particularly with people who use alternative means of communication, may not have easy access to technology and possess little technical literacy.

Socioeconomic differences in the frequency and patterns of internet use are well documented[53,54]. Thus, enhancing diversity will require the use of multilingual, multiplatform advertising the enhanced use of mixed methods for data collection and ideally, the involvement of an interdisciplinary team including social scientists, epidemiologists and qualitative research methodologists.

Data handling:

The scope of PSPs has expanded to include questions about prognosis, diagnosis and health care delivery. The volume of data collected in the uncertainty gathering phase has increased in step with the expanding scope. In our scoping review, this was especially noted in high prevalence conditions, such as acne. To make matters more difficult, a number of PSPs reported that questions were submitted as anecdotes, which some groups mined for possible research questions, whereas others discarded. Data appraisal to determine whether or not the question has already been answered by the literature, was also noted to be more difficult in our scoping review. The original JLA method was developed to prioritize uncertainties about the effect of treatments. Thus, an uncertainty was deemed to exist if a systematic review concluded the effect was not known, or if there was no systematic review addressing the question. These limited criteria for assessing whether a question remains unanswered has been difficult to extrapolate to questions about diagnosis, prognosis and health care delivery, partly because they are not typically studied using clinical trials and systematic reviews in the way treatments are. In our own MDS PSP, we did not face a problem with high volume data, in part owing to the lower incidence of MDS and the relatively low number of responses. We did, however, have a high number of anecdotal submissions which required more careful analysis to determine whether a research question could be uncovered. This certainly added to the resource intensity of the data handling portion of the PSP, as has been reported in the literature[44,64,82], but mining anecdotes for potential research questions could be an important step in developing a more inclusive engagement process.

Underscoring this growing challenge, the JLA Guidebook 2021[30] edition includes a step by step guide and special considerations for data handling and acknowledges that the methods for this phase of the process continues to evolve.

Results Usability:

Doubts about the usability and generalizability of the top 10 research priorities were commonly reported in our scoping review. These concerns were primarily related to two factors. The first is closely related to the diversity of the participants, as discussed above. A second factor however, was that because the final prioritization in the JLA method is a consensus driven process, it often results in more general questions with broader appeal being prioritized over more specific questions. More general questions require further refinement and subdivision by researchers in order for them to be truly useful for framing future research. Thus, although the broader priorities may have wider applicability, they may not be as useable in their agreed upon form as guideposts for researchers, and therefore less effective at accomplishing their intended purpose of directing future research. Our experience with the JLA process (Chapter 3) did not extend into the prioritization phase, so we are unable to draw any comparisons between our experience and those of others.

Implications:

Implementation of the JLA method:

Our work has several implications for enhancing the future implementation of the JLA approach to research priority setting. First, the issue of inclusion is central to the exercise of priority setting. The broad adoption of the JLA methodology represents a significant move towards engaging people with lived experience and point of care health care workers in determining research priorities. Broad engagement with marginalized peoples however, requires considering who these people are, what barriers they may face in engaging and addressing these barriers specifically. For example, including Indigenous peoples, people from ethnic and linguistic minorities or those in remote locations is unlikely to result from a general survey call on social media in English or French. Beyond sampling methods, the actual means through which research questions are gathered can also play a role in making the process more inclusive. Many groups found, including our own, that patients and caregivers often submitted anecdotes instead of research questions. This may point

to the fact that many people, including clinicians, may find the task of formulating an actual research question daunting, despite possessing a wealth of lived practical experience to draw upon. Thus, one way of allowing broader and more open participation is to mine submitted anecdotes for potential research questions, or to actually ask for anecdotes. The latter choice may have the added benefit of encouraging a more diverse group of people to engage with the process in the first place.

Greater inclusion and greater diversity in sampling and data gathering will make the JLA process even more resource intensive and should thus be undertaken only if the PSPs have a real desire and ability to do this. In our view, not every PSP will need to be comprehensively inclusive at every step, but every PSP should clearly document its methods and who it was able to reach so that the agreed upon "top 10" can be properly contextualized and any gaps, if present, can be addressed by other groups.

One way of balancing diversity and resource intensity of the JLA process is to re-narrow the scope of query, such that PSPs are attempting to prioritize research questions about one or two related areas of research, such as treatment and care delivery, for example, as opposed to conducting a broad PSP which attempts to be all things to all stakeholders. A narrower scope would, in theory at least, enable a greater focus on methodological rigor and may result in more usable results, although this remains unknown.

Result usability and the tension between broad and specific questions was raised by several groups included in the scoping review. Broader questions affecting a greater number of stakeholders tend to be prioritized over more specific questions, yet it is the latter types of questions that are typically answerable by individual research projects. Therefore, narrowing the scope of PSPs may enable stakeholder groups to develop more useable research questions which can then be prioritized in relation to other related questions. The agreed upon priorities are then ready to be moved on to actual study development and funding, thus enhancing the value of PSPs.

Another way of optimizing the utility of results from PSPs is to include researchers who are not connected to the specific condition being prioritized, but who have expertise in reaching diverse populations and

conducting social research, including analyzing complex qualitative data. These can include epidemiologists, social scientists and qualitative researchers who can lend their expertise to support the work of the PSP.

Policy:

A significant body of literature exists on the importance of patient engagement in research[3,5,36] and public research funding agencies and industry have adopted models for encouraging such engagement[34,35,134]. There remains, however, a paucity of data on the real-world impacts that engagement has had on health policy. This may partially be explained by the fact that the move towards patient engagement is relatively new, and a significant time lag is needed to observe and study its effects on research policy. At the same time, our scoping review raises a number of issues related to the implications of patient engagement in research prioritization. Most of the arguments in support of patient and clinician involvement in setting the research agenda centre around direct benefits, i.e. the benefit of generating more relevant research to patients themselves. Outside of the JLA context, evidence points to a strong relationship between disease advocacy and an increase in disease-specific research funding[135], whereby every 1000 dollars spent on advocacy resulted in an additional 25000 dollars in research funding. In our review, the presence of strong patient advocacy organizations is a facilitator to engagement. Thus, it would be reasonable to predict that if the research prioritization initiative is supported by a strong patient advocacy organization, the identified research priorities would be favoured in the allocation of research funding. To date, we have found no studies focused on understanding the impact of prioritization exercises on research funding. In our view, given that the first PSP was conducted over 15 years ago, proof of real world follow-through on the research priorities identified by JLA PSPs would do a great deal to enhance the value and financing of such initiatives.

On the other hand, the effectiveness of advocacy organizations in attracting research dollars towards questions important to them means that these dollars may be diverted away from conditions or questions affecting people less able to organize and fundraise, such as conditions primarily affecting marginalized

groups, who often lack the resources needed to engage in high level lobbying and advocacy work. In that respect, the JLA method is certainly not immune from bias. In our scoping review we identified the presence of strong patient advocacy organizations willing and able to act as partners as a strong factor in the success of the PSP. Taken together with the concerns about the possible lack of diversity in engagement, this points to the strong possibility that while the JLA method is a step towards inclusion of patient voices, important groups may be left behind. Seen through a broader societal lens, advocacy and patient engagement can also have potentially negative systemic effects[135], whereby greater involvement by privileged groups of people inadvertently contributes to increasing the marginalization of others. Prioritization is necessarily, a political process. Those who can and do engage in the process have their voices heard. The danger exists if their voices are taken to represent "the" voice of patients. In light of the increasing interest in participatory research, the time is right for acknowledging these important limitations and allocating funds towards studying its intended and unintended consequences on society as whole. Understanding the impact of prioritization methods such as the JLA on society as a whole and science more specifically is an important first step in deciding how to maximize their potential benefits while minimizing potential harms.

Given that many public research funding agencies have encouraged patient engagement in research, and given the possible biases in these processes, serious consideration should be given to whether public funders of research should be involved in directly funding PSPs in a manner that reduces the heavy dependence on advocacy organizations as vehicles of engagement, and ensures a minimum standard of equity and inclusion in the process. As discussed above, it may not be necessary for every PSP to include everyone, but an awareness of whose voice may be missing and how that impacts the business of knowledge creation and implementation is crucial.

Conclusions and Future Directions:

There is no doubt that the JLA approach to research prioritization is an important method which is growing in popularity. Overall, this method provides a step in the right direction towards giving people with lived experience of MDS, their caregivers, and clinicians greater influence over the research agenda. Our review

has identified several areas of uncertainty, both in the method and impact of PSPs as a vehicle for research prioritization. One question is whether re-narrowing the scope of PSPs would allow for a deeper understanding of priorities. Future research should compare the conduct and real world results of broad vs narrow scope of PSPs. Do narrower PSPs result in more usable priorities? Do they allow for deeper engagement with a more diverse group of patients and health care providers?

The other area of interest is the development of more specific tools that would facilitate PSP implementation. For example, several groups found interim prioritization to be quite challenging. Developing and testing a tool which would simplify this step would be helpful in simplifying the task for individual PSPs. Another area is the development of tools to facilitate research question formulation. These can be public facing, aimed at patients and health care providers who are not researchers, or they can be aimed at those conducting PSPs, offering a valuable educational resource on how to maximize the utility of their collected data.

One of the biggest limitations of our review is the lack of reporting on the process of conducting research prioritization using the JLA method and instead focusing on reporting the "Top 10" research priorities. In our view, the field would benefit from greater stringency in adopting reporting guidelines for JLA PSP and ensuring that others can benefit from the experiences of those who have real world experience using this important method for research prioritization.

Finally, the question underlying this body of work is whether the research priorities identified using the JLA method are being prioritized by funders or independently taken up by researchers. In our view, this question lies at the heart of whether this type of work is in fact, delivering on its promise of maximizing the value of health research.

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