The future of myeloma research in Canada and beyond: results of a James Lind Alliance priority setting partnership

Multiple myeloma, often referred to as myeloma, is an incurable cancer of the plasma cells which accumulate in the bone marrow and produce monoclonal protein resulting in hypercalcaemia, renal insufficiency, anaemia and bone deterioration. To complicate matters further, both the disease and its treatment can impair organ function, sometimes in similar ways, contributing to significant disease burden, which can impair the quality of life for those living with myeloma.

This is problematic given the growing incidence of the disease; between 1996 and 2016, the global incidence of myeloma increased by 126%. Simultaneously, research into treatment options for myeloma has led to people living longer. However, since there is currently no cure for myeloma, future research is needed to advance the diagnosis, treatment and management of myeloma and improve the quality of life of those living with this disease.

Studies have shown that the uptake of new research evidence into clinical practice is not immediate, nor direct; on the contrary, the process can take years and, even then, not all evidence becomes part of practice. One factor contributing to this gap is the lack of communication between research authors and research users. Understanding what is important to those directly affected by the condition being studied — by involving these key stakeholders prior to conducting research — is one way of bridging this divide.

One such approach is the James Lind Alliance (JLA) Priority Setting Partnership (PSP), which seeks to foster greater alignment between differing research agendas by bringing together patients, caregivers and clinicians to determine research priorities. Past PSPs have highlighted how research users and research authors often have diverging priorities: for example, alternative interventions and therapies versus drug-centred treatments. At the same time, research priorities identified through previous PSPs have often been successful in obtaining funding from government agencies or charitable foundations. This has encouraged researchers to focus on questions most important to research users through competitions and funding opportunities.

Established in 2019, our project on the future of myeloma research in Canada builds upon the successes of previous PSPs (for more information the project, visit: https://www.jla.nihr.ac.uk/priority-setting-partnerships/myeloma/). Faithful to JLA guidelines, we assembled a pan-Canadian steering group composed of people living with myeloma, caregivers and clinicians. Together, these members guided all four phases of the iterative priority setting process, which are described below. Informed consent was obtained from...
participants at each stage of the process and materials and procedures were approved by the Horizon Health Network Research Ethics Board (RS #: 2019-2788).

Among the materials developed for the project was an open-ended survey, which asked participants to submit questions that they considered important for research. This survey represented the first phase of the project. Close to 600 participants completed the survey, submitting topics of interest in each of the four identified areas: diagnosis, treatment, management and living well with myeloma. Through this initial survey, over 3,000 questions, or ‘uncertainties’, were put forward by people living with myeloma, caregivers and clinicians.

Once the survey was closed, the collected uncertainties were grouped into 76 indicative questions, and the steering group was called upon to help identify those that were within the scope of the project. Then, the indicative questions were checked against more than 600 systematic reviews and 300 clinical practice guidelines published between 2010 and 2020. The goal was to ensure that existing research had not already answered these questions. As a result, 45 of the original 76 indicative questions were brought forward to the next phase.

As in the first phase, the third phase of the project also centred on a survey, this one asking participants to choose from the provided list of 45 indicative questions and rank their personal ‘top 10’. This close-ended, interim prioritization survey, which collected over 650 responses, was used by the steering group to determine which questions (n = 18) would be moving to the final phase — the priority-setting workshop.

It was during this online workshop, held in April 2021, which a final, ranked list of the Top 10 research priorities in multiple myeloma was agreed upon (see Table 1). This consensus was arrived at by 23 patients, caregivers and healthcare providers, helped along by a group of JLA advisers who encouraged discussion from all participants and aided in the prioritization of questions using nominal group techniques.

Interestingly, the priorities identified through this process were largely related to pharmacological treatments, including a cure for the disease as well as new treatments (e.g., immunotherapies and personalized medicine) that improve quality of life, while having fewer side effects. Participants also expressed a desire for treatments for specific myeloma-related issues, such as bone deterioration, or for specific types of myeloma, such as relapsed, refractory, or drug-resistant myeloma as well as high-risk or aggressive myeloma.

The predominant focus on pharmacological treatments for myeloma was surprising given the well-documented mismatch between the priorities of research consumers and those of the research community. Indeed, one review found that pharmacological interventions accounted for 18% of priorities identified by previous PSPs. Yet, despite the broad approach taken by the project to identify research priorities for all areas of myeloma care, the majority of the Top-10 priorities were drug-related.

These results speak to both the importance of drug treatments for patients’ quality of life and the immediate need to address concerns associated with these treatments. While this emphasis on pharmacological interventions suggests that there is potential alignment between research consumers and the research community, the priorities highlighted by this study also provide funders and researchers with specific areas of focus within this broad domain.

The Canadian Myeloma PSP was the first project to bring together people living with myeloma, caregivers and clinicians to identify their shared priorities for the future of myeloma research. The insights gained from these individuals, combined with the methodological rigour and transparency that are at the very core of the JLA approach, ensure that these priorities are meaningful and relevant to members of the myeloma community.

A recent addition to the long line of multistakeholder collaborations using the JLA PSP model, this study hopes to inform the funding of myeloma research going forward, spur original research and systematic reviews focusing on these priorities and, consequently, improve the quality of life for people affected by myeloma.

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Author contributions

SF, LM and TR conceptualized the study; LM and TR acquired research funding; SF, LM, SB, MR and TR did project administration; all authors contributed to investigation and methodology; SF, LM, SB, MR and TR curated data and did formal data analysis with input from all authors; SF, SB and MR prepared the original draft manuscript; all authors reviewed and the edited manuscript; TR supervised the study.

Conflicts of interest

DR reports consultancy for and honoraria and research funding from Janssen, Celgene, a Bristol-Myers Squibb company, Amgen, Takeda, serving on board of directors or advisory committee for Janssen and Celgene, a Bristol-Myers Squibb company, honoraria and research funding from Merck and research funding from Otsuka; TR reports contracts with AstraZeneca, Roche, Pfizer, Amgen and BristolMyersSquibb; KS reports honoraria and research funding from Celgene, a Bristol-Myers Squibb company, Janssen, Amgen, Takeda; LD, DR, TR and WP are members of the Myeloma Canada Board of Directors.

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