Determining patient and carer priorities in inclusion body myositis: a patient-led research study

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Abstract

Objective

Inclusion body myositis (IBM) is a rare neuromuscular disease that confers significant functional disability. Understanding the priorities of IBM patients and carers is critical to directing care interventions where resources are limited. This Australian patient-led study aimed to identify current challenges for IBM patients and carers; prioritisation of these challenges; and self-reported ability to cope.

Methods

This phased study used a mixed methods approach. An initial qualitative 'discovery' phase, involving a small cohort of patients and carers (n=23), determined key challenges (themes). A subsequent quantification phase, involving 149 Australian IBM patients and carers, utilised an online survey to prioritise themes and determine current level of coping.

Results

The study identified 10 inter-related themes, and their relative importance was then determined. Ninety-five percent of total importance came from 7 themes: 1. uncertain future; 2. coping with daily frustrations; 3. lack of cure, treatment and understanding; 4. impact on carer's capabilities and own needs; 5. change of roles and relationships; (6) Getting information, education and support; and 7. significant impact on mental health. Other themes identified were: 8. financial impact; 9. issues with government-provided care provision; and 10. concerns around access to voluntary euthanasia. Participants reported low mean 'coping scores' across all themes.

Conclusion

This study identified gaps in care, education and support. The highest priority themes focussed on practical challenges of living with IBM in addition to significant impact on mental health. Understanding the priorities of IBM patients and carers is critical in directing resources and providing person-centered care and support.

Key words

inclusion body myositis, cost of illness, quality of life, mental health, patient participation

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Introduction

Inclusion body myositis (IBM) is a rare neuromuscular disease that causes progressive and permanent disability through autoimmune attack on skeletal muscle (1-3). Predominantly affecting people aged 50 years and over (3, 4), typical presentation includes weakness and wasting of the quadriceps, forearm muscles and finger flexors (2). Two-thirds of IBM patients experience dysphagia (5). There are currently no disease-modifying treatments available for IBM (4), and research is ongoing to improve understanding of its aetiology and pathogenesis. Diagnosis of IBM is often delayed due to the varied clinical presentations, as well as patient and health system barriers (6, 7).

The physical and functional impact of IBM is varied, aligned with heterogeny of the clinical phenotype, particularly at presentation and early in disease (1, 4, 8). However, over time there is a reliable progression towards significant functional disability related to global weakness particularly affecting quadriceps, finger flexors and bulbar muscles (5, 9). As the disease progresses, patients with IBM require increasing assistance with activities of daily living and often become reliant on carer support (4). Within 20 years of diagnosis, most patients will become wheelchair dependent (5).

The patient journey often starts many years before formal diagnosis, navigating the challenges of recognising and acknowledging symptoms, finding accessible specialist care and obtaining a definitive diagnosis (7). Beyond diagnosis, patients navigate uncertainty regarding disease severity and trajectory alongside day-to-day physical impacts. These challenges are compounded by the relative lack of community information and support due to the rarity of IBM.

Understanding the impact of IBM for patients and carers is critically important to ensure that clinical care and research includes consideration of the key issues for the patient alongside clinical and scientific imperatives. This means improving understanding of the lived experience of IBM. Although research focussed on holistic impacts of myositis is increasing (10-12), there currently remains a paucity of research focussed on understanding the specific priorities of IBM patients and their carers.

Globally, there is rapidly growing recognition of the importance of person-centred research, which includes participation and leadership of research by patients and carers themselves (13-15). Patient-led research is increasingly recognised for its importance in gaining unique insights into lived experiences and helping to understand unmet needs (16). This research study was initiated by a person living with IBM, who recognised unaddressed burden of disease within their own experience as well as in the experience of others. The study was conceptualised by this patient-researcher, who proposed to apply 'action research' techniques (more commonly found in business, marketing, and social sciences) to identify and prioritise the unmet needs for patients and carers living with IBM.

The purpose of this Australian study was to discover the priorities of IBM patients and carers as seen through their own eyes; to assess the relative importance of these priorities; and to measure patient and carer perception of their ability to cope with these challenges.

Materials and methods

The study utilised a mixed methods approach. A sequential exploratory design was chosen, with an initial qualitative ('discovery') phase to identify challenges (priorities), followed by a quantitative phase, where priorities were ranked and coping levels assessed. This study design is summarised in Figure 1.

The study was reviewed and approved by the Murdoch University Human Research Ethics Committee (Approvals 2022/118 (qualitative phase) and 2023/094 (quantitative phase)). The study was conceived by a patient-researcher partner and designed in consultation with myositis patients and carers, including members of the Myositis Research Consumer Panel. The study benefitted from significant consumer (patient) involvement, particularly in respect to participant-facing documents and the quantitative phase survey design.

The qualitative 'discovery' phase utilised action research techniques within

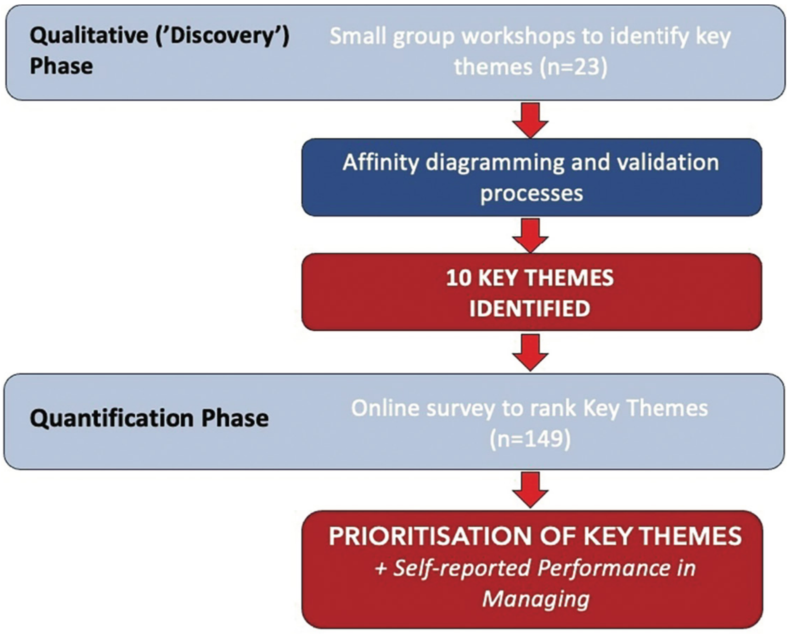


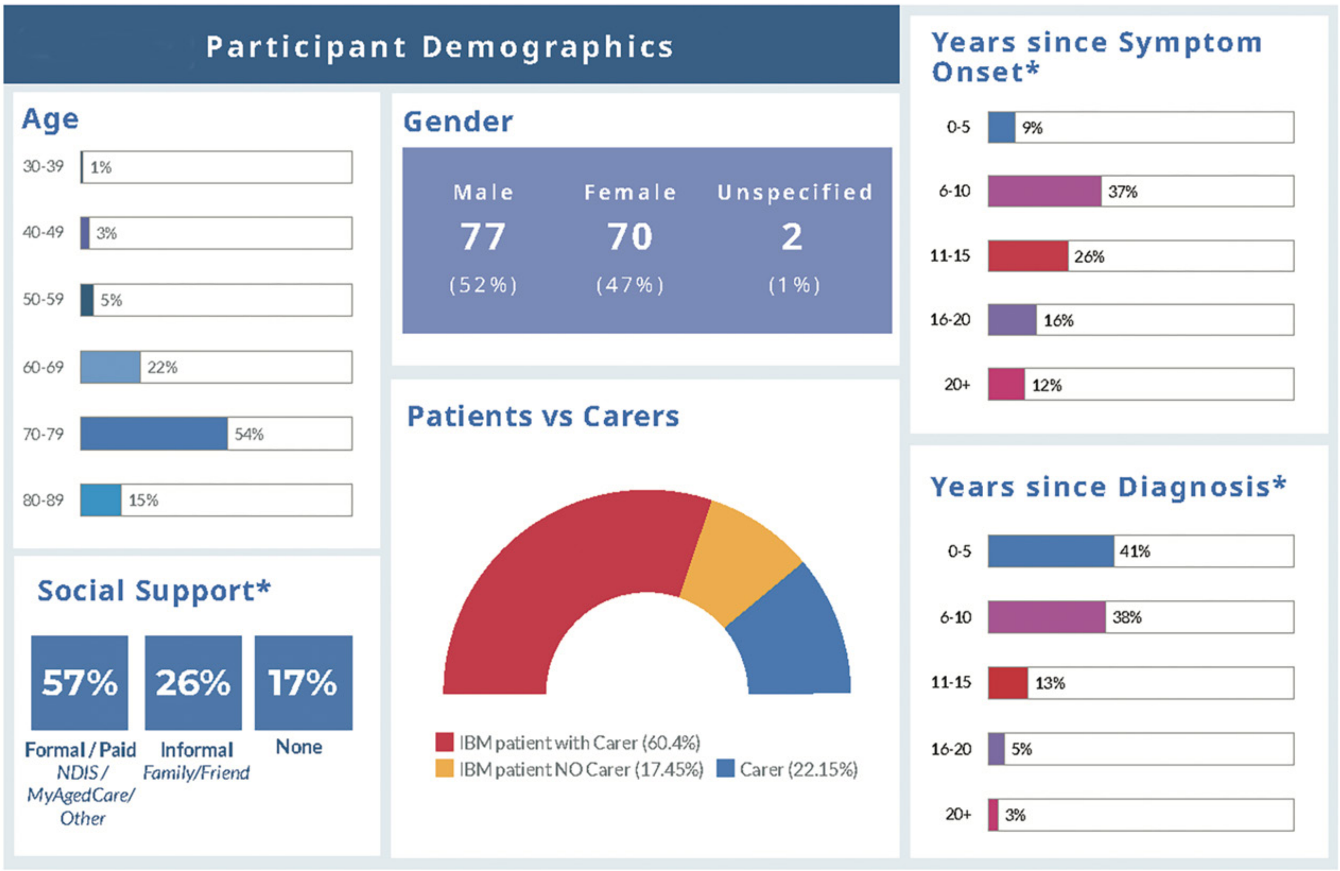
Fig. 1. Study design.

semi-structured group interviews. Action research refers to a style of research approach founded within social sciences (17), and focused around key elements of: participation and collaboration; knowledge sharing and building; processes of reflection and observation; social change and problem-solving (18, 19). Action research has a growing acceptance within healthcare research (20, 21) and presents a strong case for use as a patient-centred clinical research tool, with its integration of participant and researcher knowledge and focus on highly translatable outcomes of action and change (22). The specific approach for this study utilised materials and methods developed by Enzyme Group, a specialised action research consultancy group, with resources provided under licence and contracted support from Enzyme Group (co-author Hawkins).

The qualitative study phase was completed over a 3-month period in late 2022/early 2023 and involved 23 IBM patients and carers. Participants were recruited via email invitation from the Myositis Association of Australia, completing an online expression of interest form with basic demographic information (including sex, age range, type of myositis, duration of disease, geographical location, level of support). From the pool of interested patients and carers, responses were coded for demographic elements and computer-generated randomisation used to select participants to ensure a broad representation of demographic characteristics. Selected participants were invited to attend one of three online workshops, held in November 2022. Written, informed consent was received from all participants. During the workshop, facilitators guided participants through three sets of questions related to living with IBM, utilising aided and unaided recall techniques. Group discussion was encouraged, with experiences and perspectives shared. Participants used a workbook to record their personal brainstorming. This workbook was retained by the participant and not shared with the research team. Following the workshop, participants were asked to review their workbook to identify up to six 'priorities', using a template feedback sheet (Appendix A, online). Each participant completed two feedback sheets, one from their own perspective (*i.e.* that of a patient or carer), and the second from the alternate perspective (*i.e.* patient considering priorities of a carer, and *vice versa*). For each of the items reported on the feedback sheets, participants were asked to indicate the relative importance of each item, using a 3-Point Likert scale (Important, Very Important, Critical), and to rank their current 'performance in managing' each item, using a 10-point Visual Analogue Scale (VAS), with 0 as 'Very poor' and 10 as 'Outstanding'. Relative importance rankings were weighted logarithmically as follows: 'Important' was scored with a weight of 1; 'Very Important' was scored with a weight of 3 and a Relative Importance of 'Critical' was scored with a weight of 9. This 1-3-9 ranking system derives from quality improvement processes used within business sectors, in particular Quality Function Deployment (QFD) (23) and Six Sigma approaches (24, 25). The use of a 1-3-9 ranking system offers several benefits for prioritisation, by allowing highly ranked factors to be easily identified, creating greater distinction between high and low ranked factors, and reducing the likelihood of ties between factors (23-25).

De-identified feedback sheets were digitally transcribed and an 'affinity diagramming' technique used to organise the items into broad themes. Affinity diagramming is a commonly used approach within business and social sciences to organise ideas and data (26). This visually-led technique involves grouping related data items together and establishing a thematic focus for each group. For this study, affinity diagramming was completed within two online sessions by the study team, with a further validation session that included four study participants (2 patients and 2 carers). The affinity diagramming process resulted in 10 clear themes being identified. Headings for each theme were generated from wording used in participant responses, to ensure that theme headings reflected findings as closely as possible.

The second phase of the study involved quantification of the identified themes to elicit overall order of priorities for both patients and carers. This phase utilised an online survey, with members of the Myositis Association of Australia and the Myositis Discovery Programme (a specialist myositis research clinic in Perth, Australia) invited to take part, via an anonymised link. The invitation was sent to 512 individuals. The survey was open for 5 weeks, with two reminder emails sent during this period. Patients



*Relates to the patient, and, in the case of carers, to the diagnosis, symptoms, and social support of the patient they care for.

Fig. 2. Participant demographics.

and carers with all forms of myositis were invited to participate in the quantification phase, however, only results from IBM patients and carers are presented in this paper, as they constituted the largest and most homogeneous patient population. IBM patients and carers were identified via self-reported diagnosis data fields within the survey.

Within the online survey, the 10 themes (priorities) from the qualitative phase were ranked by each participant to elicit an overall order of priorities for patients and carers. The quantification phase utilised forced binary trade-off, asking participants to prioritise between unique pairings of the 10 themes, until all themes had been compared with each other (45 questions). Participants were asked to indicate their current 'performance in managing' (using the 0-10 VAS). For the quantification phase, participants reported only on their own perspective (*i.e.* patient or carer). Demographic data was collected, as per the qualitative phase, and participants were provided with the opportunity to add any themes that they felt had not been included. Participants were also invited to optionally contribute free text comments, to ensure that no ideas or important items were missed. The final hierarchy of priorities was calculated by placing the themes in rank order as determined by the participants' scores (both patients and carers together), taking the lowest scored theme and setting at 0, taking the top scoring theme and expressing it as 100, and then expressing all the other theme weightings as a percentage of 100. This resulted in 'relative importance' scores for all themes.

Results

Demographics

For the qualitative phase, 56 expressions of interest were received from 43 IBM patients and 13 carers. All carers were invited to take part. Patient participants were selected based on several criteria to ensure diversity and representativeness. 13 patients and 10 carers participated in the qualitative phase. For the quantitative phase, 116 IBM patients and 33 carers participated. Both phases included broad geographical spread of participants across Australian States and Territories, with similar representation of metro (56%) and regional/rural (44%) participants. A summary of participant demographics within the quantitative phase is shown in Figure 2.

Thematic discovery

Within the qualitative phase, 10 key themes were identified, as shown in Figure 3.

Relative importance

The most important theme identified within the quantitative phase was '*Uncertain future*', with the least important, '*Concern not covered by voluntary euthanasia*'. Ninety-five (95%) percent of the total weight of relative importance came from 7 of the 10 themes: *Uncertain future*', '*Coping*

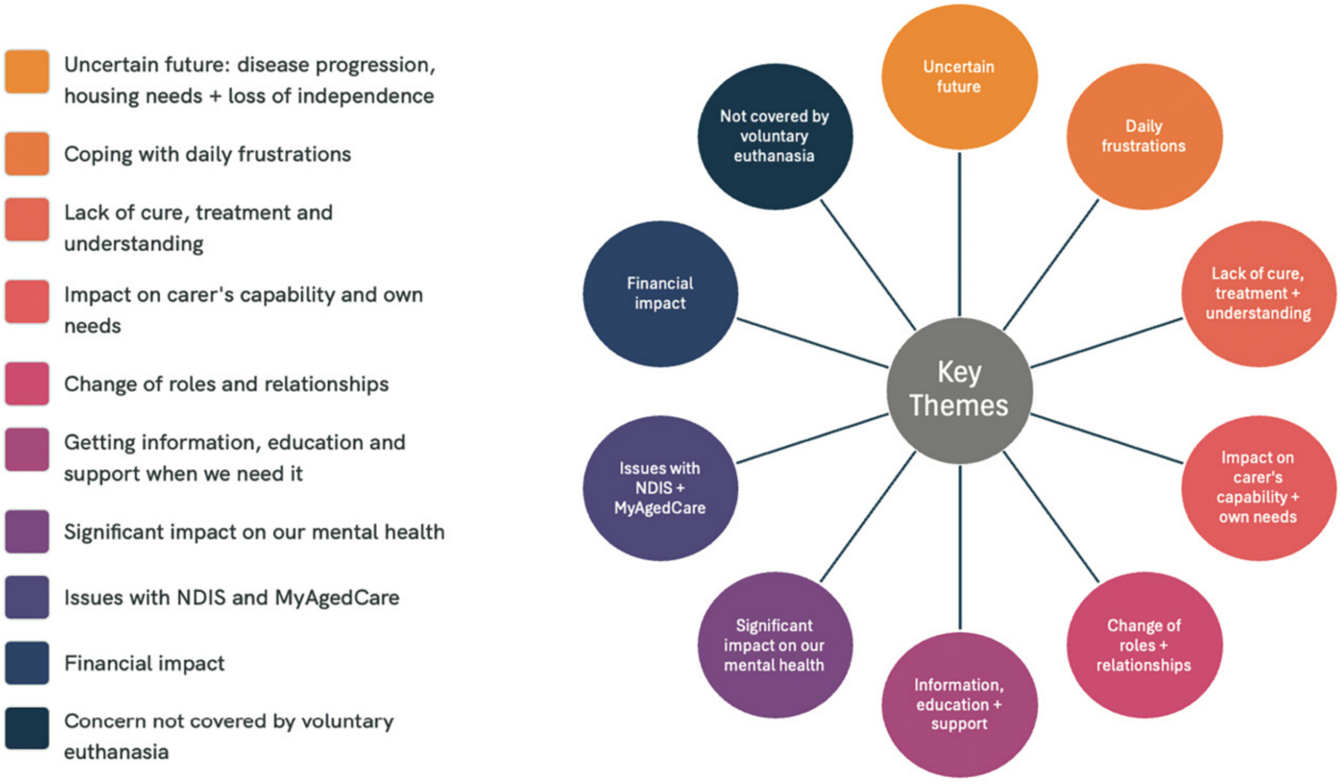


Fig. 3. Key themes.

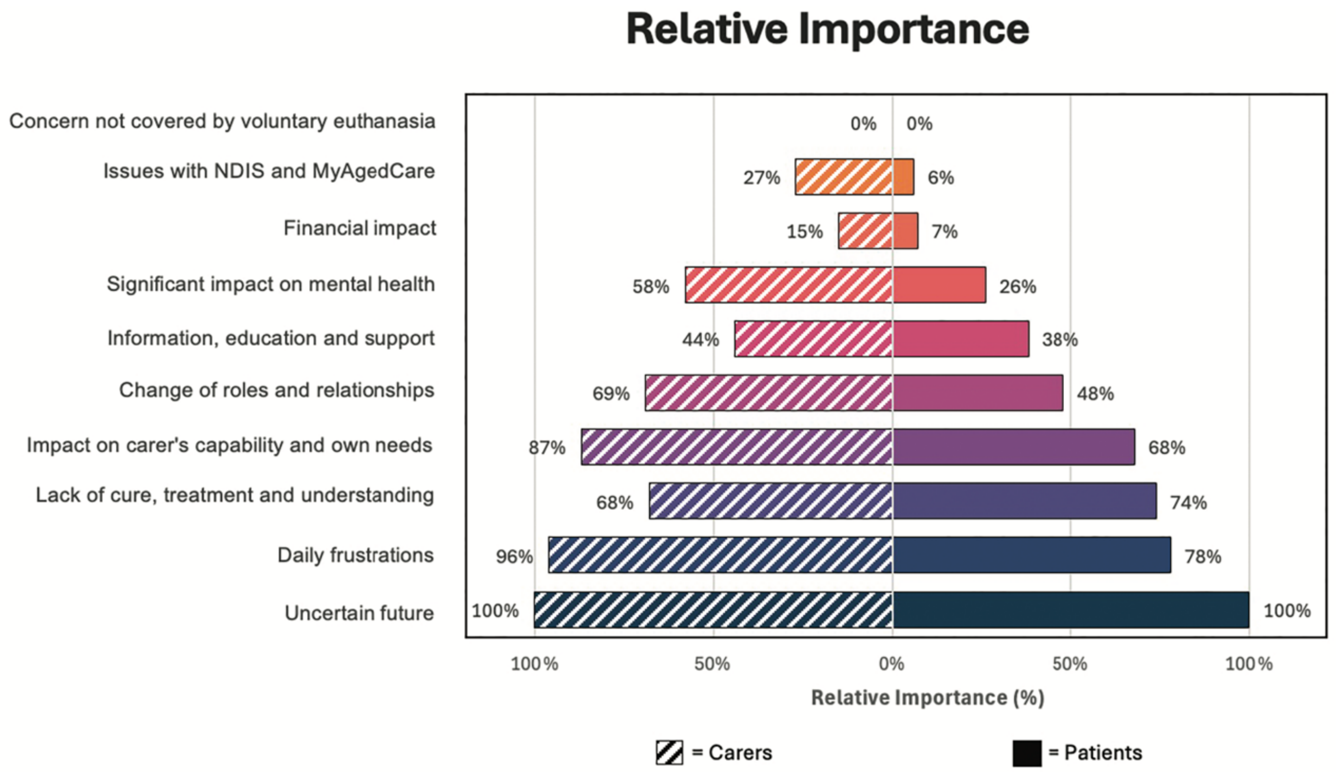


Fig. 4. Relative importance.

with daily frustration', *'Lack of cure, treatment and understanding'*, *'Impact on carer's capability and own needs'*, *'Change of roles and relationships'*, *'Getting information, education and support when we need it'*, and *'Significant impact on our mental health'*. Priorities were determined for patient and carer subgroups. A summary of the results is shown in Figure 4. Relative importance was similar for patients with carers, as well as patients without car-

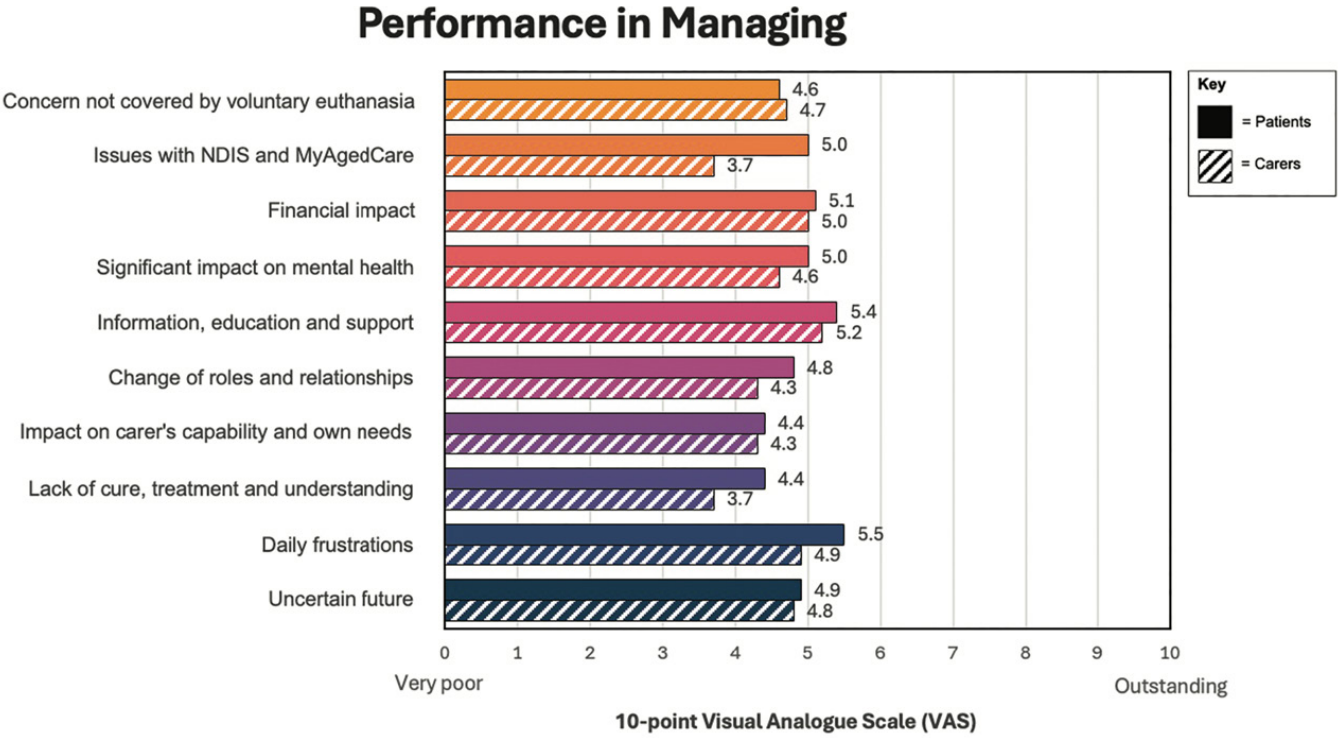


Fig. 5. Performance in managing.

ers. Similarly, no significant differences were observed between patients with formal, informal or no social support.

Performance in managing

Across both phases of the study, 'coping scores' for each theme were determined by use of functional indices calculated from the VAS results. Within the qualitative phase, mean 'coping score' was less than 60% across all themes for patients and carers. Within the quantitative phase, overall IBM patient coping score was 46.7%, and overall IBM carer coping score was 43.1%. Coping scores against each theme are shown in Figure 5.

Discussion

This patient-driven study sought to better understand the priorities of patients and their carers living day-to-day with IBM. The study identified 10 key themes focussed around practical as well as existential challenges related to living with a progressively disabling disease. When ranked, the theme of 'uncertainty', as related to future needs and abilities, was found to be most important. Other highly prioritised themes of '*Coping with daily frustrations*', and '*Lack of cure, treatment and understanding*' illustrate a sense of helplessness and a significant impact on self-determination. Our study found that patients and carers are not coping well.

Holistic priorities

Although this is not the first attempt at discovering patient priorities in myositis (8, 10, 27-32), to our knowledge this is the first study focused on patients living with IBM and their carers. It is also unique in seeking to determine panoptic priorities rather than focusing on research or clinical priorities. Within the broader myositis literature, in particular the myositis OMERACT group (33) have been working for a number of years to evaluate and establish patient-reported outcome measures relevant for myositis (with the exception of IBM) (12). This included identification of the domains considered most important to patients, using the OMERACT methodology (34). Of the 638 survey respondents in their study, the domains selected as most important included "muscle symptoms," "fatigue," "physical activity," "medication side effects," and "pain" (12).

In contrast to the findings of the OMERACT group, our study identified broader issues related to living with a chronic, progressive, untreatable disease such as IBM. Themes of "uncertain future", "daily frustrations" and "lack of treatments" were identified as the most important, highlighting key differences between people living with IBM, and other (more treatable) forms of myositis. Consistent with understanding of the phenotype of IBM (5), pain was not identified within either study phase, including within free text comments. The differences identified may also reflect the advantages of the action research approach in enabling a deeper understanding of disease impact and identifying themes beyond physical symptoms.

Within the ranking of priorities, both patients and carers assigned least importance to the theme of "*Not being covered by voluntary euthanasia*". Within Australia, each State and Territory has its own legislation regarding Voluntary Assisted Dying (VAD), with varying eligibility criteria (35). The emergence of this theme from within the qualitative phase of the study indicates its significance for this patient group. Whilst this theme was not universally prioritised, those who did select this issue as a priority within the quantification phase consistently ranked it highly, indicating the extent to which IBM is impacting some patients.

Interestingly, a recent study looking at the patient journey and health-related quality of life in 7 IBM patients (10) suggests that priorities may change throughout their journey, starting with uncertainty about physical vulnerability, then passing through a phase of promising treatments and self-management until the stage of the weak body and increased caregiver burden arises. Their corresponding informational support, physical support and emotional support requirements change during these phases. Majority of respondents in our study (79%) were ≤10 years since diagnosis, which may mean that the hierarchy of priorities reported is most reflective of a disease phase where self-management is still predominant. Further sub-group analyses of the study data are ongoing, which will consider the role of disease duration more fully.

Mental health

Our study confirmed a significant mental health impact of IBM on both patients and carers. Mental health impacts were reflected within numerous themes, from the explicit theme of "*Significant impact on our mental health*" to more pervasive impacts including within the highest ranked themes of "*Coping with daily frustrations*" and "*Uncertain future*". This reflects the increasing body of literature recognising the impact of IBM (and other chronic, untreatable conditions) on quality-of-life, including mental health (30), and the need for further research and intervention focussed in this area. The importance of mental health support for myositis patients and carers is recognised as a priority area (11, 30, 32, 36), with ongoing research to identifying appropriate screening and interventions. Certainly, the saturation of mental health-related issues identified in our study suggests that clinicians should evaluate current care of IBM patients to consider if mental health assessment and support could be improved.

Carers

A recent qualitative study of IBM patients (10) identified that a supportive patient-carer relationship may confer a positive impact on health-related quality of life, when compared to patients without supportive carers. In a disabling disease like IBM, the transition of a spouse or partner to a full-time carer can be challenging for both parties. This was identified in our study within the theme of *"Change in roles and relationships"*, which was more highly prioritised by carers (72% relative importance) than patients (48% relative importance). Interestingly, carers within our study identified the theme of *"Significant impact on our mental health"* as more important than patients, also ranking *"Coping with daily frustrations"* and *"Financial impact"* higher than patients. This indicates a significant carer burden associated with IBM despite disease stage, with 58% of the patients being cared for reported as earlier in disease (symptom onset ≤10 years) and therefore likely to have less carer support requirements than later in disease. A recent patient-led study on carer burden in myositis found that caregiver burden is positively correlated with disease duration beyond 6 years, with burden increasing as disease progresses (32).

Coping

Alongside identifying the priorities of patients and carers, this study sought to understand how well patients and carers are currently coping with those priorities, providing key information to direct interventions and support. A strength of the action research approach is the intention of uncovering tangible actions or changes that can be implemented in response to the research (19, 20).

Our study identified universally moderate-to-low self-reported 'coping' scores. Across all themes, mean coping scores ranged from 38% – 57%. Overall coping scores were very similar for patients and carers, although the average coping score of carers was lower than patients', indicating that patients are coping slightly better than carers. Carers reported the lowest coping scores within the themes of "*Issues with NDIS and MyAgedCare*" and *"Lack of treatment, cure and understanding"*, both themes that include system barriers that patients and carers are not easily able to influence. Patients reported the lowest coping scores related to *"Lack of treatment, cure and understanding"* as well as *"Impact on carer's capability and own needs"*. The use of coping scores within the study provides a useful metric that could be used to measure the success of future interventions targeted towards the priorities identified in the study.

Strengths and limitations

The study's methodology allowed for true discovery of the priorities of IBM patients and carers, beyond the limitations of pre-determined domains. The action research techniques provided an opportunity to identify key issues that were not only most meaningful, but also highly translatable into further research streams and direct actions to improve care. As a patient-led initiative, this study provides a true reflection of what is important to patients to research, and the type of research questions that we should ask.

There were several limitations that should be noted. The study only included participants from Australia, meaning that regional-specific nuances should be considered, such as access to universal health care at low or no cost (Medicare) (37). Information regarding IBM diagnosis and symptom/diagnosis date were self-reported, with information not independently verified. The demographic data fields collected were limited, with a view to minimising personal information collected, however omission of data such as ethnicity or income has limited our ability to analyse the data with these lenses and to understand if the recruitment approach was robust enough to reach under-represented individuals. Likewise, the omission of any disease severity measures (such as IBMFRS or ambulatory status) prevents analysis of responses according to disease severity. The study is also limited by the low number of carer participants relative to patients. Overall, sub-group analyses are limited by the small numbers of participants in each sub-group, meaning that the results suggest further research questions, rather than fully answering them.

Conclusion

Overall, this study helps us understand the breadth of issues that clinicians need to consider when providing holistic care to IBM patients and their carers. The study has allowed us to discover firsthand, the direct set of experiences faced by IBM patients and carers. It enables both deeper insights into some of the practical issues faced by patients, as well as mental health loads and additional challenges impacting day-to-day living. The identified themes suggest a framework for novel clinical tools, and the study team have been exploring development of a patient-reported instrument that would help to guide care and support interventions during clinical consultations. Alongside this, a working group has been formed within the Myositis Association of Australia to address the highly identified priority of mental health, producing an initial draft of strategies that offer practical translation of the study findings.

We hope that highlighting the priorities of IBM patients and carers will help everyone involved in IBM care to be cognisant of the impact of disease, allow clinicians to screen for difficulties in these domains, and advocate for improved access to care, support and improved allocation of limited resources for each patient.

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