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PRACTICAL APPLICATION

Designing Discrete Choice Experiments Using a Patient-Oriented Approach

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Abstract

Patient-oriented research is a process whereby patients or caregivers are included as research partners so that research focusses on topics that are priorities and lead to findings that translate into practice. Using a case study of preferences for stem cell transplant in scleroderma, we report on a patient-oriented research approach to developing a discrete choice experiment. Our patient-oriented research application followed the four guiding principles in Canada's Strategy for Patient-Oriented Research: inclusiveness, support, mutual respect and co-build. In this case study, patient partners were involved at different levels of engagement to match individual availability, skillset and roles in the team. They advised, to different degrees, on all aspects of the study from design to analyses. Using a patient-oriented research approach led to the inclusion of attributes that would likely have been excluded (e.g. support from a multidisciplinary team), and realistic framing of patient-relevant and sometimes sensitive attributes (e.g. mortality and cost). Meeting locations and times were adjusted to accommodate all-team circumstances. Institutional constraints on the reimbursement for patient partners influenced the timing and extent of involvement. We found that adopting a patient-oriented research approach to discrete choice experiment design injected unique knowledge and expertise into the team, improved the representativeness of the sample recruited, minimised researcher biases, and ensured appropriate attribute selection and descriptions. The patient-oriented research approach highlighted some constraints of discrete choice experiment designs and, while not a solution, might ensure the methodological trade-offs remain patient relevant. Institutional challenges must be addressed to progress patient-oriented health economics research.

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Key Points for Decision Makers

Using a patient-oriented approach to develop a discrete choice experience might contribute to greater transparency, acceptability and appropriateness of the methods, and increase participation in research.

Researchers looking to integrate patient partners in their research team need to consider that patients often join teams while keeping their full-time jobs, responsibilities and social commitments; accommodations must be made that allow inclusiveness and avoid burnout.

Institutional challenges need to be confronted to enhance how patient partners can contribute to research projects at their full potential. Involving patient partners can help to identify and highlight methodological shortcomings of research designs.

1 Introduction

Patient-oriented research (POR) is "a continuum of research that engages patients as partners, focusses on patient-identified priorities and improves patient outcomes" [1]. In the last decade, POR became the gold standard in clinical research and permeated adjacent fields, including health economics. The approach moves away from the traditional paradigms where the patient is the object of the research, to a model in which the patient's experience is seen as valuable to the co-creation of knowledge. Patient *partnership* in research is therefore distinct from patient participation in research. Through POR, patients are involved as team members to contribute to the study's design, not as research participants who provide data used to answer a research question. Patient-oriented research constitutes a significant paradigm change motivated by a recognition that moving away from paternalistic research models is ethically, politically and socially sound, particularly in publicly funded research [2]. Patient-oriented research can lead to better alignment between research designs and patient preferences, which can increase research participation, uptake and impact [1].

Using POR might also contribute to reduced waste in research. Recent estimates suggest that 85% of research funding is wasted as a result of poor reporting of results, inefficient prioritisation of research funding, poor design, conduct, and analysis, and lack of knowledge translation [3–5]. For example, studies that used discrete choice experiments (DCEs) to predict uptake of new rheumatoid arthritis preventative treatments suggest that few drugs being studied would be acceptable [6, 7]. These studies were conducted after the trials were completed, and large investments made. If a POR approach is used *prior* to clinical trials, research funds might be channelled to studying treatments that are most likely to be preferred by the people they are intended for, which would in turn lead to improved outcomes.

Discrete choice experiments are a type of survey that allows researchers to elicit stated preferences for goods or services [8]. In healthcare, DCEs are particularly useful to create hypothetical markets of health technologies that are not yet available, allowing understanding of peoples' preferences and the trade-offs they are willing to make when considering, for example, a new treatment. One of the main challenges of stated preferences methods is the potential disconnect between stated and revealed (actual) preferences, known as hypothetical bias [9]. A recent systematic review and meta-analysis of DCEs concluded that DCEs generate reasonable predictions of health-related behaviors [10].

Following methodological guidelines for DCE development can help minimise hypothetical bias [11]. Development of DCEs requires thorough understanding of the decision problem being simulated, achieved through literature reviews, stakeholder consultation and qualitative research [8, 12]. Poor design and complexity can result in inexplicable choices, hindering the analysis or interpretation of the results [13]. It is recommended that researchers pay careful attention to and adequately report the many stages of survey development, including choosing the alternatives, identifying attributes and levels, and deciding on how each component is described and worded [14]. This process offers many early opportunities for patient involvement, which can lead to higher levels of engagement throughout the remainder of the study.

In Canada, POR is now a requirement for obtaining health research funding, and it is therefore likely that more researchers, including health economists, will seek to engage patients more in their projects. This article contributes to increased transparency and harmonisation in POR by reporting on its application to the development of a DCE using a case study where we elicited the preferences of people with scleroderma for autologous hematopoietic stem cell transplant (AHSCT) [Box 1].

2 Box 1 Background information about the case study

2.1 Scleroderma

Scleroderma is a chronic autoimmune disease that causes the body's connective tissue to harden. Localised scleroderma usually affects the skin only, while systemic scleroderma, its most severe form, can affect the skin, muscles, joints, and internal organs with severe consequences including life-threatening lung and heart failure. There is currently no cure and the disease is mainly managed using immunosuppressant therapy.

2.2 Autologous Hematopoietic Stem Cell Transplant

Autologous hematopoietic stem cell transplant (AHSCT) was developed to treat cancers that affect the blood, bone marrow and lymph nodes. It has subsequently shown to have potential for treating some autoimmune diseases, and has recently been tested as a treatment for systemic scleroderma.

With AHSCT, a patient's own stem cells are collected and stored. The patient then undergoes high-dose chemotherapy and/or radiation therapy to remove defective immune cells. The stem cells are subsequently re-infused to reconstitute a healthy immune system. Autologous hematopoietic stem cell transplant carries significant risks, including treatment-related mortality, burdens, including travelling to specialised centres, and costs, both direct and indirect.

2.3 Rational for Using a Discrete Choice Experiment

Autologous hematopoietic stem cell transplant has shown promising efficacy in the few clinical trials completed [11, 12]. In one of these trials low recruitment led to the entry criteria being broadened and the primary endpoint changed to a global rank composite score to reduce the sample size required [11]. The difficulty in recruitment suggests that not all people with scleroderma and eligible for AHSCT will undergo this treatment. Furthermore, it is unclear whether the global rank composite score is meaningful to people with scleroderma considering this treatment. It is important to understand the factors (attributes) that will influence patients' preferences for AHSCT and the acceptability of treatment, and how these would affect the uptake of treatment in clinical trial settings and in routine care. Discrete choice experiments are an established approach to obtaining this information.

2.4 The Discrete Choice Experiment

We designed a discrete choice experiment that asked participants to choose between two AHSCT alternatives, or no AHSCT treatment (opt-out). The selected attributes were:

- Years after treatment without further scleroderma organ damage;
- immune suppression treatment and risk of immediate complications;
- late complications (i.e. cancer);
- additional members to your care (in addition to your rheumatologist);
- number of people with scleroderma the hematologist has treated using a stem cell transplant;
- additional cost to you (expenses not covered by the provincial health plan, nor your health insurance);
- distance of treatment centre to your home.

Besides the discrete choice experiment, the questionnaire included three additional sections: one asked about the respondent demographic characteristics, type of scleroderma and how the disease has affected them financially; another presented basic information about scleroderma and AHSCT and asked respondents a few basic questions about the information provided, and a final section asked about the respondent's quality of life.

3 Approach

Based on the four guiding principles for integrating patient engagement into research—inclusiveness, support, mutual respect and co-build—established by The Patient Engagement Framework [1], we report on how we applied a POR approach to develop a DCE.

3.1 Inclusiveness

> "Patient engagement in research integrates a diversity of patient perspectives and research is reflective of their contribution – i.e., patients are bringing their lives into this." [1]

We recruited two patient partners, TB and JB, to our team that also included clinical experts, health economists, health systems researchers and knowledge translation specialists. Patient partners' roles in the team were discussed and defined according to their availability to commit, experience, interest in being part of a research study and health. TB and JB contributed lived experienced with scleroderma, knowledge of the patient community, and clinical environment in British Columbia, and experience in participating in discussion groups with stakeholders, including patients. The research team met regularly to make decisions as the project progressed, including decisions about planning of research activities such as recruitment, informational materials, survey development, survey administration, data collection, interpretation and knowledge translation.

3.2 Support

> "Adequate support and flexibility are provided to patient participants to ensure that they can contribute fully to discussions and decisions. This implies creating safe environments that promote honest interactions, cultural competence, training, and education. Support also implies financial compensation for their involvement." [1]

Initial meetings with the patient partners covered the environment the project was set in, and what sort of accommodations could be made to help the patient partners feel integrated. Patient partners were asked to record the number of hours they dedicated to the study and were reimbursed for their time. Time was dedicated to communicate methodological concepts and share materials so that equal participation in methodological decisions could be achieved. Opportunities were identified for patients to participate in talks and conferences.

3.3 Mutual Respect

> "Researchers, practitioners and patients acknowledge and value each other's expertise and experiential knowledge." [1]

The study was developed by a multidisciplinary team in recognition that each party would bring valuable skills and knowledge to the project. Early meetings were set up for the members to familiarise with each other's background, skills and experiences. An open dialogue about needs and expectations was kept throughout the project. A practical example of mutual respect was the effort to, whenever possible, send materials to the patient partners and allowing time to review and contribute to consequent decisions, even if this meant readjusting timelines. Decisions were made as a team, and methodological choices discussed and negotiated. It was suggested that the patient partners kept a log about their experience for separate reporting in conferences, journals and policy communications; a patient partner reflection was submitted as a commentary to this issue [15].

3.4 Co-build

> "Patients, researchers and practitioners work together from the beginning to identify problems and gaps, set priorities for research and work together to produce and implement solutions." [1]

The idea for this project was conceived by TB (patient partner) and MHa (health economist) through a conversation at a meeting on AHSCT in Montreal 2018. A research proposal was jointly prepared and subsequently funded by the Health Economics and Decision Modelling cluster of the British Columbia Support Unit [16]. The patient partners were involved in methodological decisions, data collection, interpretation, attribute selection and survey design. Following existing classification for the level of engagement [17] we aimed, in the least, to engage the patient partners at the 'Collaborate' level.

4 Application

A summary of how the POR approach was implemented in the different stages of the research project is presented in Table 1.

4.1 Inclusiveness

We achieved different levels of engagement with each patient partner and at different stages of the project (Table 1). TB was involved at a collaborative level and JB at a consultative level. We worked with our patient partners to achieve a manageable level of engagement that they felt comfortable with. Flexibility to accept the level of engagement each party could offer was important.

4.2 Support

In recognition of the patient partners' commitments (full-time jobs, volunteering work and managing their health), the team sought to accommodate their schedules and minimise travelling. Team meetings were held outside working hours and off-campus, with the option to join in via a phone call, especially as JB was not local to the team. Patient partners were supported to independently communicate their experience in this project. TB presented her experience at the International Shared Decision Making conference (2019), participated in local events such as the Health Economics and Simulation Modelling academic half-day and contributed with a commentary on her experience of working with us on this project to accompany this paper in the special issue [15]. Reimbursement for time spent at these events was offered, in recognition that participation at conferences required use of vacation time, as well as usual expenses.

4.3 Mutual Respect

Mutual respect was a goal throughout the project and resulted in recognition of the value of each team member's perspective. Patient partners' views were considered in decisions and incorporated whenever possible. Mutual respect was fundamental for decisions where compromises had to be made to assure methodological feasibility. For example, patient partners felt that chemotherapy was an important attribute in itself and should be separated from the description of other possible complications. Such input was considered and several iterations of the attribute list were proposed until patients agreed with the final set. Complications of AHSCT were ultimately described using two attributes, one for short-term complications including death from the treatment and the other for long-term complications including cancer. These trade-offs between methodological limitations and patient preferences were discussed to ensure that they were not seen as a devaluation of the patients' contribution.

4.4 Co-build

TB was involved in the research project from the beginning, including the pre-funding stages of formulating the research question and writing the grant proposal. TB was also involved in preparing the focus groups material, facilitating the focus group, data interpretation and identification of attributes. From the beginning, TB highlighted the value of recruiting a second patient partner with a different lived experience, and supported the team in establishing contact

Table 1 Description of the level of involvement of the patient partners based on the IAP2 spectrum of public participation [17], example of the patient partners' contribution and the resulting differences to the methods for each step of the discrete choice experiment (DCE) development [18]

Step in DCE development [18]	Overall level of involvement [17]	Example of how patient partner contribution changed the study	Differences in the methods led by POR
Research question	Co-creation	Idea for the research project, the hypothesis and the perspective were formulated with the patient partner (TB)	The evidence gap that this study intends to fill was highlighted by the patient partner. The project would not have been conceived if the patient partner and members of the research team did not meet at a clinical event and discuss the topic
Data collection	Involve	Patient partners provided input on focus groups design and sampling and participated in the focus group, TB as a co-facilitator and JB as an observer, via remote focus group software	The patient partners highlighted the importance of including focus group participants that live outside the lower mainland, which led to an adaptation of the nominal group technique used in the focus groups to include in-person and remote participation. Improved power dynamics during qualitative data collection owing to the presence of patients as members of the research team. Both patient partners attended the focus group and co-facilitated the discussion with a social scientist, which was helpful to the next step—identification of attributes and levels
Identification of preliminary attributes and levels	Empower	The selection of the preliminary list of attributes and levels was entirely led by the patients that participated in the focus groups, which was co-facilitated by the patient partners. To enable this, we used the nominal group technique to give all participants the opportunity to communicate all the things that matter when choosing a new treatment such as AHSCT	There was only minimal intervention of the researcher facilitator in the focus group discussion. Using the nominal group technique, the participants were asked to think about the umbrella question: "What factors would matter to you if you were considering a stem cell transplant for the treatment of scleroderma". In a round robin style, each had a chance to share their ideas. The ideas were then clarified and discussed as a group, and a list of attributes and levels was created by combining similar ideas and ranking them
Selecting and defining final list of attributes and levels	Collaboration	The qualitative analysis was performed by MA and TL and a report of the findings sent to the whole team. A meeting with the patient partners was set up to discuss the attributes identified, the wording of the attributes and the levels that described them	Following the patient partners' input, the cost and distance attributes' levels were defined as a range instead of finite values; mortality was included as one of the risks of AHSCT procedures. The wording of the attribute 'years after treatment without further scleroderma organ damage' was fully derived by the patient partners. Patient patterns suggested a longer list of attributes than what MA and TL reported from the focus group qualitative data analysis, which led to a thorough discussion about the attribute and potential interactions. For example, distance of treatment centre from home (included) and time spent away from home (excluded). It led to focused literature reviews for AHSCT in the area of cancer treatment to see how attributes have been chosen and depicted in DCEs
Construction of tasks and experimental design	Consulting	The patient partners considered the survey long, which prompted discussions about the trade-offs between survey length and total number of attributes and levels	A blocked design was chosen to accommodate a longer list of attributes without increasing the length of the survey
Instrument design	Collaboration	The patient partners suggested using the survey to share with patient community current evidence-based knowledge about AHSCT to treat scleroderma. The patients noted that discussion groups were very active with the risk of misinformation propagating	A new section was introduced before the DCE in a format that shared evidence-based information and supported respondents interpreting the attributes and the choice scenarios presented. After each block of information, a quick multi-choice question was asked to confirm that the information had been assimilated. Responses to these questions will also be used to test survey validity

Table 1 (continued)

Step in DCE development [18]	Overall level of involvement [17]	Example of how patient partner contribution changed the study	Differences in the methods led by POR
Data collection: DCE	Collaboration	Recruitment strategy was discussed and planned with input from the patient partners. TB supported the dissemination of a paper version of the survey, as well as the link to the online survey through the Scleroderma Association of BC	Inclusion of a paper version of the survey for those who attended a patient event in Vancouver. Enhanced recruitment throughout BC and Canada through local and national scleroderma associations
Results and conclusions (ongoing)	Collaboration	We aim to meet as a team to discuss the survey results, and rely on patient partners' knowledge to draw conclusions. We will open discussion for the formulation of new research questions based on the DCE results, as well as the additional data collected in the survey	We will seek patient partner input, as well as extended consultation of other patients in BC, to identify further research questions based on the data collection and results of the survey
Knowledge translation (ongoing)	Empower	The patient partner contributed to the planning of the knowledge translation activities since the early stages of developing the project proposal. TB presented at the ISDM Society, wrote a commentary about their experience as a patient partner, and contributed to a presentation at the academic-half day of the Health Economics and Simulation Modelling Methods Cluster in Vancouver	The team has made strong efforts to promote the voice of the patient partners and their experiences, being positive or negative, of working in this research project. TB presented at the 10th ISDM Conference (2019), leading the knowledge translation component

DCE development steps follow as per Bridges et al. [18]

AHSCT autologous hematopoietic stem cell transplant, *ISDM* International Shared Decision Making, *POR* patient-oriented research

with JB, our second patient partner. JB joined in time to participate in the focus group via a conference call, as an observer, and provided consulting on focus groups materials, data interpretation, identification of attributes and levels, survey design and piloting.

Patient partner involvement with the design of the formative qualitative data collection stage was decisive to the direction the project took (Table 1). They recognised that our design for focus groups, to be conducted in person in Vancouver, would exclude the perspectives of those living in rural and remote areas. Furthermore, these participants would likely have different perspectives and factors influencing their decision making. We therefore enabled participants living outside the metro Vancouver area to participate via a conference call. TB took the role of a co-facilitator in the focus group, which aimed to ease power dynamics, build trust, and assist in translating concepts between the researchers and focus group participants. JB's participation as an observer in the focus group proved helpful for debriefing and interpretation of the results.

The patient partners had a central role in attribute selection and on how attributes should be worded and the choices framed (Table 1). For example, they were helpful in wording sensitive attributes for patients, such as mortality and chemotherapy-associated risks, which researchers were uncomfortable with. Patient partners stressed the importance of including non-clinical attributes, such as 'support from multidisciplinary team' and the 'distance of the treatment from your home', which were also ranked as important by focus groups participants. Following the patient partners' interest in using the survey to share evidence-based information about AHSCT, we added a set of pre-DCE questions providing information about this novel treatment and testing the respondents' knowledge about the information given. This was a way to share evidence, provide context for the attributes and levels, and test respondents' understanding of those attributes before they started the DCE.

5 Discussion

In this article, we report on the application of a POR approach to developing a DCE following the patient engagement framework [1]. Overall, we found that this approach brought value to the research project. In line with suggestions for POR initiatives, we were able to formulate a research question that aligned with patients' priorities and conducted the work in recognition of the patients' values, which resulted in a rewarding experience for the team. Having patients as members of the research team increased the perceived trustworthiness of the project to participants, which helped with recruitment and dissemination, as reported elsewhere [19]. A POR approach also created a more reflective and critical atmosphere regarding research, knowledge and power within the study team, which likely would not have been the case without the inclusion of patient partners.

The patient partners were engaged at two different levels: collaboration (TB) and consultation (JB). It seems important that those involved in POR, including researchers, supporters and funding agencies, accept that more engagement is not necessarily better [20] and not to preclude potential patient partners from getting involved solely to achieve the highest level of engagement. As tools to evaluate POR and its impact become available [21, 22], consideration must be given to the expected impact of different engagement levels.

This team corroborates other researchers' experiences that POR can be time and resource intensive [23, 24]. Inclusiveness meant recognising that patient partners are active members of society with ongoing commitments including full-time jobs, volunteer work, caring responsibilities and their health condition. Project planning and budget estimations must accommodate these, as well as patient partners' training, support for travelling, flexibility in scheduling meetings and other activities.

We also found that POR approaches might collide with principles of research governance. Engagement with patient partners ideally starts as early as possible, before funding applications are submitted, to allow, for example, for the co-formulation of the research questions and study design. This means that patients are asked to contribute long before the project is funded and approved by an ethics committee board, i.e. they are involved for a longer period than that of the project's timeline. During this time, researchers might not have a way of compensating patients for their time and expertise. Given that not all projects are successful in funding competitions, patient partners are at risk of becoming professionally, financially and emotionally invested in projects that might not be taken forward.

There were some challenges in articulating a POR approach within a DCE study. For example, patient partners would have preferred that more attributes were included while also voicing concerns that the survey itself was too long [12 choice sets in total, and completion time of around 30 min]. The patient partners were also concerned that the hypothetical treatment options might be misinterpreted as actual options. Many of these issues are known methodological challenges for DCEs. For instance, Coast et al. describe tensions in developing DCEs, particularly in narrowing down rich qualitative data into a finite manageable number of attributes [12]. The number of attributes to include in a DCE is contended, with recommendations to limit attributes to assure cognitive feasibility and statistical efficiency, but including all relevant attributes to limit omitted attribute bias [8, 25, 26]. Though research in this latter area exists, an ideal number has never been suggested. Thus, researchers

tend to deal with this issue on a case-by-case basis. Explaining and justifying the methods to the patient partners was a critical step because it built trust and a common ground of understanding to promote everyone's participation in such methodological decisions. While we were not able to accommodate some of the patients' preferences, taking a POR approach ensured the omitted attributes were accounted for in the scenario descriptions, the chances of misinterpretation of the scenarios were minimised and the included attributes were the most relevant to patients.

Patient-oriented research can be met with resistance within the research community. One of the reasons might be that it can be wrongly perceived as a type of qualitative research, and its value measured using the same rigorous standards expected from qualitative research methods. Whilst qualitative research collects data to answer a research question following rigorous theoretical frameworks [27], POR aims to draw on patients' expertise to guide key decisions about aspects of the research project. This explains why in a POR approach, it is neither possible nor the aim to guarantee representativeness and inclusion of diverse views. Another reason for resistance to POR might be that patients' contributions to the direction of a project might be interpreted externally as advocacy. In Canada, training is available for patient partners that covers research governance, ethics and research methods. It needs to be recognised that in POR, patients should have the same status that any member of a multidisciplinary team would have. Therefore, the patients' contrasting viewpoints, goals or opinions should not be viewed any differently than those of the health economist, the clinician or the statistician in the team.

Power imbalances are an inherent issue in POR and one that can reduce the level of patient involvement to a minimum if preventive measures are not in place [28]. Yet, academia is built on rooted power structures, notably power of knowledge, that conflict with POR's endeavour to empower patients to influence research [29]. Patient partners often occupy a more vulnerable position than other team members. Strategies to involve patients in the research process and mitigate power imbalances rely on the researchers' skills to dialogue, convene and communicate, as well as the creation of a safe environment [30]. Currently, the patient partner integration and compensation in Canada amounts to that of a volunteer position. With the requirement by many funding agencies to incorporate POR into project proposals, the opportunity and space for tokenism is inevitably created. Patient-oriented research guidelines and best practices are needed to ensure that POR is incorporated into research projects in ways that mitigate some of the issues we highlighted and to prevent POR from becoming another possible source of waste of valuable resources in the research process.

6 Conclusions and Future Directions

Patient-oriented research might be a way to overcome challenges that, for many years, researchers have struggled with: ensuring transparency, accountability, acceptability and appropriateness of the methods, increasing participation in research, taking diverse perspectives into account and reducing waste by increasing the uptake of research findings into practice. Applying a POR approach to the DCE design can embed a unique knowledge and expertise that may affect methodological choices and enhance the scope and meaningfulness of health economics research. In addition to evaluating the impact of POR on research outcomes, institutional challenges and some methodological shortcomings of DCE need to be confronted to move such a rewarding research paradigm forward.

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Author Contributions MH, TB and TL conceived the idea for this study. NB was involved in further refinement of the research plan and provided guidance on using patient-oriented research to discrete choice experiments. SM provided expertise on patient-oriented research approaches to qualitative research and knowledge translation. MA and MH led the process of conducting all research activities. All co-authors supported the research activities' design and implementation. MA wrote the first draft of the manuscript. All co-authors reviewed, provided critical revisions for and approved the final version of the manuscript.

Compliance with Ethical Standards

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