

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	“Adding another spinning plate to an already busy life”. Benefits and risks in patient partner-researcher relationships: a qualitative study of patient partners’ experiences in a Canadian health research setting
AUTHORS	Leese, Jenny; Macdonald, Graham; Kerr, Sheila; Gulka, Lianne; Hoens, Alison; Lum, Wendy; Tran, Bao Chau; Townsend, Anne; Li, Linda

VERSION 1 – REVIEW

REVIEWER	Stephanie R Morain Baylor College of Medicine, Center for Medical Ethics & Health Policy
REVIEW RETURNED	05-Mar-2018

GENERAL COMMENTS	<p>First, I'd suggest the authors consider the potential role of educational status. They note that 59% of their interviewees had at least 1 university degree. This is markedly higher than general educational trends in Canada, for which I believe only about 25% of the general population has a similar educational background. I'd have appreciated at least a brief address of this within the discussion section (e.g., is this representative of patient partners in research in general? It wouldn't be surprising if those with higher educational attainment tended to be more involved in research. Nevertheless, it does raise questions about representativeness. It may also suggest further areas for exploration as research increasingly moves towards inclusion of patients in research decision-making, if even those with relatively high levels of educational attainment struggle with "being heard" in discussions with researchers.)</p> <p>Second, I would suggest a bit more precision regarding what types of "benefits and risks" from research are being explored. The focus of the authors seems to be on psychological benefits and risks. However, at times other risks are acknowledged, including risks to the quality of the research, and potentially risks to health from overextending oneself, which could be exacerbated by the time demands of serving as a patient partner in research. I would have appreciated bit more clarity about these different types of benefits-risks, as well as a signal from the authors about how these might be weighed against one another.</p>
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REVIEWER	David L. Hahn Department of Family Medicine & Community Health, University of Wisconsin School of Medicine & Public Health, Madison, Wisconsin, USA
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REVIEW RETURNED	24-Mar-2018
GENERAL COMMENTS	<p>Title:</p> <p>“Adding another spinning plate...”</p> <p>Description:</p> <p>This is a report of qualitative interviews with a convenience sample of past and present members of a Canadian arthritis patient advisory board, aimed at thematic analysis to better understand benefits and risks in patient partner-researcher relationships.</p> <p>General Comments:</p> <p>The current trend towards meaningful patient engagement in medical research is as much a cultural as a scientific transformation. Qualitative studies incorporating patient voices are an important source of vital information.</p> <p>Strengths:</p> <p>Strategies to enhance genuine engagement are an important topic. This paper was well written and relevant. The patient quotes were compelling and supported the three derived themes (“Being Heard,” “Co-building social relationships,” and “Adding another spinning plate...”). The theme “Being Heard” nicely encapsulates and confirms other work on genuine patient engagement. “Co-building social relationships” is an important reminder that researchers and patients must give something of themselves outside of their purely professional roles, and resonates with my personal experience as a clinician and a researcher who engages patients in my work. “Adding another spinning plate...” was, for me, a novel presentation of a concept, and broadened my awareness from, for example, just thinking about the “nuts and bolts” - of convenient time, place, transportation and child care – to the broader domain of nurturing a safe environment for patients experiencing multiple demands on their time and energy. It is not just clinicians and researchers who deal with competing demands!</p> <p>Weaknesses:</p> <p>Other than the usual caveats about generalizability, I found little to criticize.</p> <p>Specific Comments:</p> <p>Title</p> <p>No concerns. I found the title “catchy” and it reflects an important point of the article.</p> <p>Abstract</p> <p>No comments</p> <p>Introduction</p> <p>Well done overall There were a couple of discipline-specific terms (“embodiment” and “foreground” as a verb) that some readers may</p>

	<p>wonder at. The former term was defined by example soon after its first use; “foreground” was not so well defined.</p> <p>Methods</p> <p>No major concerns. For the uninitiated it would be helpful to briefly describe or give an example of “constant comparative methods” in addition to referencing the method (page 7 line 51).</p> <p>Results</p> <p>No major concerns. Excellent set of quotes. Review the attribution for the footnote on page 9, line 52. Page 12 line 49: perhaps some examples here (e.g., in my experience ice breakers at working meetings, dinners, and social outings have all proved beneficial). I agree that “first name basis sans degree” is important.</p> <p>Discussion</p> <p>Page 18, lines 22 ff: It might be relevant to mention the emerging acceptance within the biomedical community of patient-reported outcome measures (PROMs) and patient-reported experience measures (PREMs). I appreciated the concluding questions re risks and benefits broadly applied (to researchers as well as to patient partners). There is more to be learned. This paper is an important step along the way.</p>
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REVIEWER	Kathryn Sibley University of Manitoba, Canada
REVIEW RETURNED	05-Apr-2018

GENERAL COMMENTS	<p>This study explored the perceptions of individuals who had been involved as patient research partners in the conduct of various health research studies through their role on a research advisory board. A relational ethics framework was used to analyze the findings.</p> <p>Overall the manuscript was well-written and easy to follow. My comments may provide some additional consideration to strengthen the paper.</p> <p>Given the three main themes identified in the findings and equal weighting given to them in reporting, it wasn't clear to me why the “spinning plate” theme was emphasized by being featured in the title. I would suggest either changing the title to better reflect the message of the results and discussion, or re-working the text sessions to emphasize the importance of the spinning plate them.</p> <p>While the focus of the study is on understanding patient partner experiences in research involvement and the authors indicated that the study was co-designed and conducted by patient partners who are co-authors, no detail is provided on HOW this was done. The reader has no way of confirming patient contributions or quality or impact of the approach. While a full analysis of the effects of the co-creation model are beyond the scope of the paper, more detail on what was done is needed to validate the reporting of co-design.</p> <p>Further elaboration of the relational ethics perspective would be helpful for readers who are not familiar with it. Where did originate? In what contexts has it been applied? What other theoretical</p>
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	<p>approaches were considered for this study and why was this one chosen?</p> <p>Did the two authors who conducted all interviews do so together or separately? Is there any researcher/ patient partner significance to the choice of individuals who conducted the interviews.</p> <p>Why did you stop interviewing at 22 of the 33 members? It is noted that a sample of convenience was used. No comment is made about saturation of themes. Please address these.</p> <p>With regards to the reporting of themes, on page 13 and 14 quotes describing positive examples of the role of two-way dialogue between patient partners and researchers are provided. I wonder if any contrasting quotes can be provided to further demonstrate this point.</p> <p>No study limitations are discussed. It is important to acknowledge that all studies have limitations. Please address this.</p> <p>I noted in the interview guide that there were several specific probes in question 2 about negative aspects of research involvement. I didn't see much discussion of this in the manuscript- did this not come up? I think these are just as important to discuss as facilitators.</p> <p>With regards to the COREQ reporting checklist, I think it would be important to address #5 (researcher experience and training- currently not reported), #6 (relationship established- says reported on page 6 but relationship between researchers, patient partners, and participants not clear to me), #7 (participant knowledge of interviewer- currently not reported), #8 (interviewer characteristics- not reported), #13 (non-participation-- # refusals not actually specified, were all 33 invited?), #17 (was interview guide piloted?), #22 (saturation not discussed).</p>
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VERSION 1 – AUTHOR RESPONSE

REVIEWER 1 COMMENTS TO THE AUTHOR:

1. I'd suggest the authors consider the potential role of educational status. They note that 59% of their interviewees had at least 1 university degree. This is markedly higher than general educational trends in Canada, for which I believe only about 25% of the general population has a similar educational background. I'd have appreciated at least a brief address of this within the discussion section (e.g., is this representative of patient partners in research in general? It wouldn't be surprising if those with higher educational attainment tended to be more involved in research. Nevertheless, it does raise questions about representativeness. It may also suggest further areas for exploration as research increasingly moves towards inclusion of patients in research decision-making, if even those with relatively high levels of attainment struggle with "being heard" in discussions with researchers).

Response: We have now addressed high levels of educational attainment among our participants, with a suggestion for further exploration of issues of diversity in patient engagement in research. Please see Page 21 (first paragraph).

2. I would suggest a bit more precision regarding what types of “benefits and risks” from research are being explored. The focus of the authors seems to be on psychological benefits and risks. However, at times other risks are acknowledged, including risks to the quality of the research, and potentially risks to health from overextending oneself, which could be exacerbated by the time demands of serving as a patient partner in research. I would have appreciated bit more clarity about these different types of benefits-risks, as well as a signal from the authors about how these might be weighed against one another?

Response: Our study examines benefits and risks that have been encountered in patient partner-researcher relationships by patient partners. The different benefits and risks we identify in our findings are grounded in the experiences of the patient partners we interviewed. Our analysis first organizes these benefits and risks into 3 thematic categories, providing insight into benefits and risks related to 1) Being Heard, 2) Co-Building Social Relations, and 3) Adding another spinning plate to an already busy life.

Next, our analysis considered these 3 thematic categories in light of ethical tenets from the relational ethics framework. We found that benefits and risks experienced by participants across thematic categories were particularly relevant to 3 ethical tenets (respect, embodiment and interdependency). Respect underscores benefits and risks related to power and vulnerability. Embodiment highlights benefits and risks related to an individual’s subjective feelings. Interdependency highlights benefits and risks that an individual experiences as part of a community, connected with other individuals and institutions making demands on their time/attention/freedom of choice.

A clearer explanation of how our analysis has organized benefits and risks experienced by participants in relation to key ethical tenets of the relational ethics framework is now provided on Page 5-6 (last paragraph) and Page 19 (first paragraph).

REVIEWER 2 COMMENTS TO THE AUTHOR:

General Comments:

1. The current trend towards meaningful patient engagement in medical research is as much a cultural as a scientific transformation. Qualitative studies incorporating patient voices are an important source of vital information.

Strengths: Strategies to enhance genuine engagement are an important topic. This paper was well written and relevant. The patient quotes were compelling and supported the three derived themes (“Being Heard,” “Co-building social relationships,” and “Adding another spinning plate...”). The theme “Being Heard” nicely encapsulates and confirms other work on genuine patient engagement. “Co-building social relationships” is an important reminder that researchers and patients must give something of themselves outside of their purely professional roles, and resonates with my personal experience as a clinician and a researcher who engages patients in my work. “Adding another spinning plate...” was, for me, a novel presentation of a concept, and broadened my awareness from, for example, just thinking about the “nuts and bolts” - of convenient time, place, transportation and child care – to the broader domain of nurturing a safe environment for patients experiencing multiple demands on their time and energy. It is not just clinicians and researchers who deal with competing demands!

2. Weaknesses: Other than the usual caveats about generalizability, I found little to criticize.

Specific Comments:

3. Title: No concerns. I found the title “catchy” and it reflects an important point of the article.
4. Abstract: No comments

Response: We thank the reviewer for the above comments.

5. Introduction: Well done overall. There were a couple of discipline-specific terms (“embodiment” and “foreground” as a verb) that some readers may wonder at. The former term was defined by example soon after its first use; “foreground” was not so well defined.

Response: We have replaced the verb ‘foreground’ to ‘highlight’ for clarity. Please see pages 5-6.

6. Methods: No major concerns. For the uninitiated it would be helpful to briefly describe or give an example of “constant comparative methods” in addition to referencing the method (page 7 line 51).

Response: We have added a brief description of constant comparative methods on Page 8 (last paragraph) and added an additional reference to support further reading about the method.

7. Page 18, lines 22 ff: It might be relevant to mention the emerging acceptance within the biomedical community of patient-reported outcomes (PROMs) and patient-reported experience measures (PREMs). I appreciated the concluding questions re risks and benefits broadly applied (to researchers as well as to patient partners). There is more to be learned. This paper is an important step along the way.

Response: We have added mention of the emerging acceptance within the biomedical community of patient report outcomes. Please see Page 19 (last paragraph).

REVIEWER 3 COMMENTS TO THE AUTHOR:

1. Given the three main themes identified in the findings and equal weighting given to them in reporting, it wasn’t clear to me why the “spinning plate” theme was emphasized by being featured in the title. I would suggest either changing the title to better reflect the message of the results and discussion, or re-working the text sessions to emphasize the importance of the spinning plate.

Response: We have revised text on Page 19 (second paragraph) to emphasize the importance of the “Spinning Plate” theme. As Reviewer 2 states, whereas the “Being Heard’ and “Co-building social relations” themes confirm other work on genuine patient engagement, the “Spinning plate” theme provides new insight because it broadens awareness of patients juggling their engagement in research with other priorities in their daily lives.

2. While the focus of the study is on understanding patient partner experiences in research involvement and the authors indicated that the study was co-designed and conducted by patient partners who are co-authors, no detail is provided on HOW this was done. The reader has no way of confirming patient contributions or quality or impact of the approach. While a full analysis of the effects of the co-creation model are beyond the scope of the paper, more detail on what was done is needed to validate the reporting of co-design.

Response: We have added a figure to provide more detail on how the study was co-designed and conducted by co-authors. The figure was created using a framework developed by Hamilton et al. to advance the reporting of patient engagement in rheumatology research. Please see Figure 1.

3. Further elaboration of the relational ethics perspective would be helpful for readers who are not familiar with it. Where did it originate? In what contexts has it been applied? What other theoretical approaches were considered for this study and why was this one chosen?

Response: We have elaborated on the relational ethics perspective as requested. Please see Page 5 (second paragraph).

4. Did the two authors who conducted all interviews do so together or separately? Is there any researcher/patient partner significance to the choice of individuals who conducted the interviews?

Response: We have clarified that two authors with training in qualitative research conducted 11 interviews each. These author also observed each other's interviews to support their ongoing training in qualitative training, if permission was granted by the participant. Please see Page 8 (first paragraph).

5. Why did you stop interviewing at 22 of the 33 members? It is noted that a sample of convenience was used. No comment is made about saturation of themes. Please address these.

Response: Of the 33 eligible members that were invited to take part in this study, a total of 22 members consented to participate. We have now added explanation that this sample of 22 consenting participants was sufficiently relevant and varied to generate rich data that addressed the aim of our study. Saturation was not used as a criterion to determine sample size because we did not aim to provide a complete description of all aspects of our phenomenon of study. Taking an exploratory approach, we present selected patterns relevant for our study's aim that provide new insight to contribute to current understanding on our phenomenon of interest. Please see Page 7 (second paragraph) and Page 9 (last paragraph).

6. With regards to the reporting of themes, on page 13 and 14 quotes describing positive examples of the role of two-way dialogue between patient partners and researchers are provided. I wonder if any contrasting quotes can be provided to further demonstrate this point.

Response: We have expanded and added quotes describing the role of two-way dialogue between patient partners and researchers to include contrasting experiences. Please see Page 15.

7. No study limitations are discussed. It is important to acknowledge that all studies have limitations. Please address this.

Response: Limitations of the study are now presented under the subheading 'Limitations' on Page 21.

8. I noted in the interview guide that there were several specific probes in question 2 about negative aspects of research involvement. I didn't see much discussion of this in the manuscript- did this not come up? I think these are just as important to discuss as facilitators.

Response: We appreciate the reviewer's feedback. Negative aspects of involvement related to the aim of this paper included instances where researchers' expertise had been discounted to the detriment of the quality of the research, negative physical and emotional impacts (e.g., feelings of inadequacy, guilt, stress, fatigue) and negative impacts for other valued relationship in participant' lives. Negative aspects are described in the results section on Pages 10-16, and in our discussion on Pages 19-20.

9. With regards to the COREQ reporting checklist, I think it would be important to address #5 (researcher experience and training- currently not reported), #6 (relationship established- says reported on page 6 but relationship between researchers, patient partners, and participants not clear to me), #7 (participant knowledge of interviewer- currently not reported), #8 (interviewer characteristics- not reported), #13 (non-participation-- # refusals not actually specified, were all 33 invited?), #17 (was interview guide piloted?), #22 (saturation not discussed).

Response:

#5) Researcher experience and training

We have now reported that researchers generating and analyzing data were doctoral students with training in qualitative research (see Page 8, first paragraph).

#6) Relationship established

We also now clarify that patient partners and participants are members of Arthritis Research Canada's Arthritis Patient Advisory Board. The Advisory Board has over a decade-long history of partnering in research with researchers at Arthritis Research Canada. The study thus builds on a pre-existing relationship among all co-authors. Participants have also engaged to varying degrees with co-authors (including researchers and patient partners) in previous research studies at Arthritis Research Canada. Participants are familiar with research interests of the interviewers. (See Page 6, last paragraph and Page 7, second paragraph).

#7) Participant knowledge of interviewer

We clarify that participants have a pre-existing relationship with interviewers, and were familiar with their research interests before consenting to participate (see Page 8, first paragraph).

#8) Interviewer characteristics

We add the interviews were not the interviewers' first encounter with the subject area of patient engagement in research (see Page 8, first paragraph).

#13) Non-Participation

We clarify that 22 of an invited 33 members of the Advisory Board participated in the study (see Page 9, last paragraph).

#17) Interview guide

We recognize the limitation that our interview guide was not piloted. Acting as representatives for our intended sample, patient partners did, however, inform the development of the interview guide by suggesting modifications and verifying that the revised interview guide was relevant to the research topic, easy to understand and suitable for prompting spontaneous descriptions of participants' experiences and feelings (see Page 7 last paragraph, and Figure 1).

#22) Data saturation

Saturation was not used as a criterion to determine sample size. Of the 33 eligible members that were invited to take part in this study, a total of 22 members consented to participate. We have included explanation that this was a sufficiently relevant and varied sample to generate rich data that

addressed the aim of our study. Our aim was to present selected patterns that provide new insight to contribute to current understanding of patient partner-researcher relationships, rather than to provide a complete description of all aspects of this phenomenon of study. Please Page 7 (second paragraph) and Page 9 (last paragraph).

We have revised and re-submitted the COREQ checklist accordingly.

We would like to thank you and the reviewers for your consideration of our manuscript. We feel the reviewers' comments have greatly helped to improve the manuscript, and hope that our responses above fully address these comments. If you have any further questions or suggestions, please do not hesitate to contact us. We look forward to your reply.

VERSION 2 – REVIEW

REVIEWER	Stephanie R Morain Baylor College of Medicine, USA
REVIEW RETURNED	29-May-2018
GENERAL COMMENTS	I thank the authors for their work in revising the manuscript. I have no further comments.
REVIEWER	David Hahn University of Wisconsin School of Medicine and Public Health, United States
REVIEW RETURNED	05-Jun-2018
GENERAL COMMENTS	I am satisfied that the author's have addressed my concerns. My only comment is that data are plural in the "Strengths and Limitations of this Study" second bullet point.
REVIEWER	Kathryn Sibley University of Manitoba, Canada
REVIEW RETURNED	11-Jun-2018
GENERAL COMMENTS	Overall I found this manuscript to be a pleasure to read and feel the revisions have greatly strengthened the manuscript. Thank you to the authors for incorporating the reviewer feedback. I have only a few outstanding comments. 1. I appreciate the increased description of relational ethics. However, in the introductory paragraph, the authors refer to it as a "lens", a "perspective", and a "framework". What exactly is it? There are meaningful differences between these terms. Please clarify and be consistent. 2. Thank you for clarifying the role of patient partners as co-investigators. I did note that they are members of the same advisory board as the participant sample. Where some co-authors also participants? If so this should be transparently reported. I suggest consulting Bowen et al Int J Health Policy Manag 2017, 6(1), 27–42 for an example of reporting when authors are also participants.

VERSION 2 – AUTHOR RESPONSE

REVIEWER 1 COMMENTS TO THE AUTHOR:

1. I thank the authors for their work in revising the manuscript. I have no further comments.

Response: We thank Dr. Morain for her review of our revisions.

REVIEWER 2 COMMENTS TO THE AUTHOR:

1. I am satisfied the authors have addressed my concerns. My only comment is that data are plural in the 'Strengths and Limitations of this Study' second bullet points.

Response: We thank Dr. Hahn for his review of our revisions and have corrected the second bullet point. Please see Page 3.

REVIEWER 3 COMMENTS TO THE AUTHOR:

Overall, I found this manuscript to be a pleasure to read and feel the revisions have greatly strengthened the manuscript. Thank you to the authors for incorporating the reviewer feedback. I have only a few outstanding comments.

1. I appreciate the increased description of relational ethics. However, in the introductory paragraph, the authors refer to it as a "lens", a "perspective" and a "framework". What exactly is it? There are meaningful differences between these terms. Please clarify and be consistent.

Response: We thank Dr. Sibley for this important comment. We have revised our description of relational ethics to clarify it as a lens. Please see consistent use of the term "lens" on Pages 2, 5, 6, 9, 19, and 21. Relational ethics stems from criticism directed towards the abstract character of much bioethical theory. It is an adaption of the theoretical grounding of bioethics to more fully reflect key components of moral life, including social and institutional relationships that inform patient decision making. It emphasizes the importance of these relational networks that are overlooked within a principle-oriented bioethical framework. The search for a more satisfactory moral grounding for bioethics has taken scholars of relational ethics in several directions. While some have dispensed with principles entirely, others believe a framework that incorporates universal principles can and should continue to constitute an adequate bioethical theory, providing these principles reflect the relational context of individual lives. Scholars of relational ethics therefore speak in different voices, reflecting different academic positions. Nonetheless, they share significant commonalities, both in their criticism of bioethics and efforts to call attention to the relational networks seldom represented within mainstream bioethics. Given there is no singular relational ethics framework, it is our belief that "lens" most accurately reflects the diversity of approaches in ongoing scholarly work on relational ethics.

2. Thank you for clarifying the role of patient partners as co-investigators. I did note that they are members of the same advisory board as the participant sample. Where some co-authors also participants? If so, this should be transparently reported. I suggest consulting Bowen et al *Int J Health Policy Manag* 2017, 6(1), 27-42 for an example of reporting when authors are also participants.

Response: Having consulted the example provided, we have added clarification that some co-authors were also participants. Please see Page 7.

We would like to thank you and the reviewers again for your review of our manuscript. We hope that our responses above fully address the most recent comments. If you have any further questions or suggestions, please do not hesitate to contact us. We look forward to your reply.