

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Involving patient research partners has a significant impact on outcomes research: an example from the international OMERACT conferences.
AUTHORS	Wit, Maarten; Abma, Tineke; Koelewijn-van Loon, Marije; Collins, Sarah; Kirwan, John

VERSION 1 - REVIEW

REVIEWER	Sherine E. Gabriel, M.D. M.Sc. William J. and Charles H. Mayo Professor Professor of Medicine and Epidemiology Mayo Clinic, USA
REVIEW RETURNED	23-Nov-2012

THE STUDY	This manuscript is largely a report of 10 years of patient engagement in OMERACT. It is not a study per se, but an account of the facts around this important initiative gathered through interviews. It is an important paper to publish but unusual so the above questions are less relevant.
GENERAL COMMENTS	This manuscript is largely a report of 10 years of patient engagement in OMERACT, with data gathered and summarizing through a structured series of interviews. The report is ground breaking in that it represents the first time a research organization has systematically involved patients in research design and the creation of core sets. This report, I suspect, will be a reference for many other organizations seeking to do the same.

REVIEWER	Laure Gossec, MD, PhD Maître de Conférence Universitaire - Praticien Hospitalier Université Pierre et Marie Curie Hôpital Pitié-Salpêtrière Service de Rhumatologie Pavillon Benjamin Delessert 2e étage
REVIEW RETURNED	18-Dec-2012

THE STUDY	Qualitative study on patient research partners
REPORTING & ETHICS	no consent or approval but not needed I think
GENERAL COMMENTS	I have read with interest this article which reports, based on literature review and qualitative interviews, the impact of patient research partner involvement on outcomes research using the case of OMERACT. The article is interesting and well-written, and indeed this is a very 'trendy' subject on which few data are available. My only problem with the article is the 'tone' which I find a bit too positive (see major comment 1).

	<p>Major comments</p> <p>1. General comment – main concern. Overall the article is a little bit TOO positive about patient involvement. When one publishes on the efficacy of a new drug, one tries to be moderate when commending the drug, and one puts forward limitations and possible draw-backs. I think the same attitude should be used here – please try to be factual, not over-enthusiastic. In some parts the article reads like an advocacy document. For example the conclusion of the abstract might be rephrased, as well as page 15 ‘enthusiastic’ etc.</p> <p>2. References There is a circularity in the references, ie the authors cite themselves a lot – although they are not the only ones to have published on this subject. Please make sure to cite other authors as well and perhaps take out some circular references.</p> <p>3. Introduction I am not sure that the paragraph about ‘development of core sets might...’ is in keeping with the article and its objectives?</p> <p>4. Methods page 5 Is the distribution across countries of interviewees representative of the OMERACT participants and if not, why?</p> <p>5. Methods/results for document analysis To present this more as a systematic review, is it possible to present the N articles searched initially, the N read as full text, and how many were used in the analysis?</p> <p>6. Length I feel the manuscript is a bit long and sometimes a little repetitive, it would be good to shorten it a bit – eg on fatigue page 11-12 as this manuscript is about patient partners not fatigue. Furthermore re. fatigue there is some amalgamation between OMERACT and the Bristol team – who also have patient partners. As this manuscript is about OMERACT patient partners I suggest to take out discussion items relevant to the Bristol work.</p> <p>Minor comments</p> <p>7. Please replace ref 18 by a website.</p> <p>8. Methods page 5 2nd paragraph: were physicians also informed of the study objective and if so, how?</p> <p>9. Ref 38 seems wrong?</p> <p>10. A ref is lacking after ‘recommended outcome’ page 11.</p> <p>11. Ref 53 and 75 is incomplete.</p> <p>12. Assumption only moving hurts page 14: this is not true in inflammatory disease anyway.</p>
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REVIEWER	<p>Professor David Evans Faculty of Health and Life Sciences University of the West of England, Bristol UK</p> <p>In terms of competing interests, one of the authors (John Kirwan) is on the steering group of an initiative I lead to support patient and public involvement in the West of England.</p>
REVIEW RETURNED	28-Dec-2012

THE STUDY	This is an important paper that provides valuable new data on the impact of public involvement in research, and I would like to see it published; however I think the methods section will need significant
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	<p>revision before it will be suitable for publication. There are several issues I believe need to be addressed. First, more methodological detail is needed on the approach to documentary analysis adopted. From the Results section it appears at times to be a historical narrative based on the authors' insider knowledge and informed by the documents, more than a systematic analysis of the documents.</p> <p>Second, as the interview topics are stated to have been derived from the documentary analysis, it seems problematic to me to claim triangulation, and if the authors wish to maintain this claim then I think it needs further explanation and justification.</p> <p>Third, there are some problematic aspects of the claimed approach to sampling. It is unclear for example, how maximum variation was achieved on the stated criteria, for example opinion about patient involvement. It is clear that a couple of participants were identified because of previous views known to the authors, but it is unclear the extent to which the authors had systematic data on all of these criteria across the potential sample. There is also a reference to adding more participants to achieve 'saturation' but it is not at all clear what this means or how it was assessed.</p> <p>Finally and most importantly, it is not until the Discussion section that the reader learns of the insider status of the researchers. This is crucial knowledge which I think should be included in the Methods section so that the reader can assess the data and analysis that follow in this light.</p>
<p>RESULTS & CONCLUSIONS</p>	<p>I have some concerns about the presentation of both the documentary analysis and interview data. In both cases, it appears to me that there is more general discussion and less clear presentation of the data than I would expect. Within the documentary analysis results section, it is sometimes hard to be sure if the discussion arises directly from the documentary data or whether it may sometimes reflect the insider knowledge of the authors (e.g. 'The chair of the conference felt confident ...' p7 52).</p> <p>With the interview results section, some sub-sections (e.g. Contributions to the research agenda) are relatively sparse with interview data compared to the amount of discussion.</p> <p>A final point concerns the discussion of the authors' insider status and the limitations of the study (pp 19-21). It is good the authors identify the potential impact of their insider status, but I think they do not give sufficient weight to the potential impact on interviewees. They state that 'no signals were identified to suggest that interviewees have simply given desirable answers' but it is equally plausible that interviewees have held back some thoughts because they knew they were being interviewed by a patient researcher and advocate of patient involvement. Some further critical reflection on the strengths and weaknesses of such insider research might be beneficial here.</p>
<p>REPORTING & ETHICS</p>	<p>There is no mention of whether the authors submitted their research for ethical review and whether approval was given. Although mention is made of a one page announcement in the patient pack, there is no indication that the interviewer and participants went through a process to ensure informed consent. Although this study may have been considered in health services terms as an evaluation rather than research and so depending on their institutional requirements may not have required a health services research ethics committee review, I would still expect it to have required</p>

	university ethics committee review. At the very least, the authors need to demonstrate a consideration of research ethics.
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VERSION 1 – AUTHOR RESPONSE

Reviewer 1

This manuscript is largely a report of 10 years of patient engagement in OMERACT. It is not a study per se, but an account of the facts around this important initiative gathered through a structured series of interviews.

response:

This is an empirical study evaluating the impact of patient participation in OMERACT conferences over the last decade. It is a systematic account of the outcomes of this process obtained through a combined analysis of interview and document data. Our findings confirm that researchers do rarely report the impact of patient involvement in their manuscripts. We therefore filled in the blind spots in the document analysis by using data derived from the interviews. To a limited extent our study does indeed present “oral history”. This is inevitable for the reason given above, but we also believe that the added value of using different resources to address our research question determines the reviewer’s qualification “groundbreaking”. Beneath we will justify that this approach represents an appropriate methodology to answer our research question.

Reviewer 2

1. My main concern is: the article is overall a little bit TOO positive about patient involvement. Try to be more moderate, more factual, and pay more attention to limitations and possible draw-backs.

response:

We believe that our research has demonstrated that patient participation has had a significant impact on the process and outcomes of OMERACT conferences according to the participants and that our manuscript provides a fair account of this process. However, to address the major comment of the reviewer we have critically assessed the abstract and the main manuscript, and reformulated our findings more moderate where appropriate. Sweeping statements and terms have been adjusted. Any generalization beyond the OMERACT conference has been removed. As suggested by the reviewer, we have also extensively elaborated on the “limitations and draw-backs” of the study in the discussion.

2. There is a circularity in the references, ie the authors cite themselves a lot – although they are not the only ones to have published on this subject. Please make sure to cite other authors as well and perhaps take out some circular references.

response:

We have tried to get a better balance in the references given by including other references related to the influence of patient participants on the development of OMERACT core-sets and outcome measures. It is inevitable to cite the authors of this manuscript when describing the relevant publications on patient participation in OMERACT conferences. For a long period the last author (JK) was responsible for the official proceedings of the patient perspective workshop and also closely involved - as an important contributor - in the research on fatigue. It is important to demonstrate here that identifying and prioritizing a new topic is not enough to influence the research agenda. As mentioned in our manuscript, by going back to OMERACT 3 (1996) where fatigue was identified by (only) professionals as an important outcome without any consequence at all (no publications about fatigue in OMERACT proceedings or other journals after 1996), it emphasizes the importance of patients raising the issue again at OMERACT 6. By referring to the relevant publication, the reader is

able to see how a topic is raised at a scientific conference, how researchers have taken the topic further on national levels and how the topic has returned on every subsequent OMERACT conference since 2002. We find it important to show that patients have been part of this process, not only at the conference, but also in the conduct of studies between conferences.

3. I am not sure that the paragraph about 'development of core sets might...' in the introduction is in keeping with the article and its objectives?

response:

Patients have been actively involved in the development of core outcome measurement sets. The development of core sets is one of the main objectives of OMERACT. For this reason it is important to briefly clarify the importance of core sets for clinical research. In the result section we have shortened the paragraph about the core-sets.

4. Is the distribution across countries of interviewees representative of the OMERACT participants and if not, why?

response:

We are convinced that the distribution over countries of the respondents is a fair representation of the total population of OMERACT delegates. Table 1 provides the characteristics of the sample in more detail.

5. To present this more as a systematic review, is it possible to present the N articles searched initially, the N read as full text, and how many were used in the analysis?

response:

In the text we justify and clarify in more detail our approach of the content analysis of relevant documents on the topic of patient participation in OMERACT and describe the scope of the analysis. See also our response to comment 2 of reviewer 3.

6. I feel the manuscript is a bit long and sometimes a little repetitive, it would be good to shorten it a bit – eg on fatigue page 11-12 as this manuscript is about patient partners not fatigue. Furthermore re. fatigue there is some amalgamation between OMERACT and the Bristol team. As this manuscript is about OMERACT patient partners I suggest to take out discussion items relevant to the Bristol work

response:

We want to emphasize that the topic of this paper is the impact of patient participation in OMERACT and we found – based on the document analysis and the findings from our interviews – that fatigue is of utmost importance in the light of the impact of patients on the research agenda of OMERACT and how this topic has been taken forward by researchers outside OMERACT. Interviewees have mentioned other topics of interest from a patient perspective, but none of these with a similar impact on the research agenda and resulting in a similar number of publications in the OMERACT conference proceedings as well as in other scientific journals. We therefore believe that including fatigue as a case-study to demonstrate how patients can influence research is justified. As a consequence, it is inevitable to include the research work done by the Bristol group that have been published in the OMERACT conference proceedings. See also our response under 2.

7. Minor comments

response:

We have corrected the reference list and addressed the other minor comments.

Reviewer 3

1. More methodological detail is needed on the approach to documentary analysis adopted. From the Results section it appears at times to be a historical narrative based on the authors' insider knowledge and informed by the documents, more than a systematic analysis of the documents.

response:

We would like to refer to our reply to reviewer 1. In addition, this study is positioned within a long tradition of hermeneutic research. In the revised method section we have comprehensively clarified the approach of the document analysis, the method of responsive evaluation, the emergent sampling strategy and the assessment of saturation and triangulation and other quality procedures common in qualitative evaluation research.

2. As the interview topics are stated to have been derived from the documentary analysis, it seems problematic to me to claim triangulation, and if the authors wish to maintain this claim then I think it needs further explanation and justification.

response:

In the 'data analysis' paragraph we explain the two meanings of triangulation that we have applied in this study: 1) Verifying findings against another source or another method; 2) As a means to enrich the data-collection and improve the face-validity by synthesizing findings from different sources. The interview topics were not only derived from the document analysis because this provided a limited overview of relevant items. We therefore held four pilot interviews and added the topic list with questions that we perceived as important to include in this study, based on our experience with working with patients in the past.

3. There are some problematic aspects of the claimed approach to sampling. It is unclear for example, how maximum variation was achieved on the stated criteria, for example opinion about patient involvement. Had the authors systematic data on all of these criteria across the potential sample? There is also a reference to adding more participants to achieve 'saturation' but it is not at all clear what this means or how it was assessed.

response:

In the 'selection of interviewees' paragraph we clarify the approach of emergent sampling and highlight how the criteria for sampling have been applied in our study. We address in particular the assessment of the criterion 'opinion about patient involvement' in relation to the authors' insights in the participants. Saturation is explained as a team responsibility to assess the robustness of the empirical data to support and describe the identified themes presented in this manuscript.

4. Most importantly, the insider status of the researchers is crucial knowledge which should be included in the methods section.

response:

We fully agree with the reviewer that this should be explained earlier in the manuscript. We now critically discuss the problems related to bias and over-identification in the method and discussion section, and how the team tried to avoid these. We hope that this revision addresses all the questions that were brought forward.

5. The presentation of both the documentary analysis and interview data: Be more clear whether findings arise directly from the documentary data or whether it reflects the insider knowledge of the authors (e.g. 'The chair of the conference felt confident ...' p7 | 52).

response:

This study has followed a methodology of responsive evaluation and reports the findings as the result of a combined review of the document analysis and the interview data. Both parts of the results section are based on this combined review. We have tried to be more explicit in acknowledging the source of particular findings: the document analysis, interview data or personal interpretations. Where appropriate the recollections of the authors have been incorporated in the text, although this has been done rarely.

We have removed the distinction between 'document analysis' and 'interviews'.

The statement of "the chair of the conference..." is directly derived from one of the interview transcripts and does not reflect any insider knowledge of the authors.

6. With the interview results section, some sub-sections (e.g. Contributions to the research agenda) are relatively sparse with interview data compared to the amount of discussion.

response:

This is correct. It can be explained by the fact that in particular the influence of patients on the research agenda is best documented by the OMERACT proceedings of the patient perspective workshop. Therefore this paragraph contains a lot of references. The influence can also be demonstrated by the use of quotes from interviewees and this has certainly steered our summary here. For the sake of the length of the manuscript we propose to stick to the current two quotes (researchers) in the text.

7. Critical reflection on the strengths and weaknesses of the insider research status might be beneficial for the discussion.

response:

In the discussion section we have added a reflection on the advantages and draw-backs of the prior engagement of two authors in OMERACT. See also our response to comment 1 of reviewer 2.

8. The authors need to demonstrate a consideration of research ethics.

In the Netherlands ethical approval is not needed if no medical data is collected and when there are no invasive interventions. We have included a statement on research ethics accordingly.

VERSION 2 – REVIEW

REVIEWER	David Evans Professor in Health Services Research (Public Involvement) University of the West of England UK No competing interests
REVIEW RETURNED	24-Mar-2013

GENERAL COMMENTS	The authors have addressed all the points I made in my first review either in amending the text or in reasonable comments in the response letter to the editors. I believe this valuable paper is now almost ready for publication. I have only two final and minor points I would ask the authors to consider. First, in four places (pp 18 and 22) they refer to equality between patients and professionals. While it is clear that in OMERACT patient and professionals have 'equal voting rights' (p 10) it is not clear that they have 'equal partnerships' or are 'full and equal collaborators'. Given the power inequalities between patients and professionals it would be surprising if this were
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	<p>the case, and I don't think the paper provides evidence for these claims, so some amendment or qualification might be appropriate.</p> <p>Second, I'm not sure if a word is missing at an important point on p22 line 16 - 'Many participants, not only partners, but also young researchers and other new-comers, are able to identify their own contribution ...' I was not sure, but I thought from the context that there might be a 'not' missing before 'able to identify'.</p>
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VERSION 2 – AUTHOR RESPONSE

The first comment:

“... in four places (pp 18 and 22) they refer to equality between patients and professionals. While it is clear that in OMERACT patient and professionals have 'equal voting rights' (p 10) it is not clear that they have 'equal partnerships' or are 'full and equal collaborators'. Given the power inequalities between patients and professionals it would be surprising if this were the case, and I don't think the paper provides evidence for these claims, so some amendment or qualification might be appropriate.”

This comment has been addressed by adding to the discussion paragraph on page 22/3. One instance referred to by the referee was a quote. However, the general point is well made and we agree that it is worth drawing attention to and discussing the distinction between two areas, while arguing that our data show evidence of full and equal collaboration, at least to some extent. We have added:

“It should be noted that striving toward equality is a normative ideal,⁸³ and fighting inequalities between patients and professionals is and remains an ongoing concern. “Equality” may be seen in two ways: as the formal position of patients at the conference (as full delegates they had the same voting rights as professional delegates, they received the same pre-conference materials and had access to all conference sessions like all other participants) and as equality of partnership or collaboration in terms of influence on the decision making process. Given the power inequalities between patients and professionals the latter is the greater challenge, but nevertheless some of the documents and interviews suggest it has been achieved to some extent. Although patients remain only indirectly represented in the executive committee (the highest decision making body), our data support the conclusion that a small number of experienced patients achieved an equal relationship with researchers in their area of interest. They obtained the competences that enabled them to perform all kinds of tasks at the conference similar to professionals, and provided input that justified co-authorship of peer-reviewed articles. We did not obtain in-depth information about the question to what extent power inequalities between patients and researchers still persist but we know from the feedback of all respondents, including some fellows and researchers who attended OMERACT for the first time, that some did not feel treated equally. To what extent this experience was caused by their status of being a patient or by the status of a new participant is still unknown.”

The second comment refers to an omission in the manuscript on our part, the word “not” should indeed be inserted in the text on page 22.