Author's response to reviews

Title: Patient engagement in Research: A Systematic Review

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Author's response to reviews: see over
Dear Editor,

We thank you and the reviewers for the time, effort and excellent advice that we followed and will hopefully make this manuscript more explicit and helpful to the readership of the journal.

Our responses to the reviewers’ comments are listed below.

MH Murad and co-authors
Reviewer's report
Reviewer: Gro Jamtvedt

Comment
In this paper the authors summaries information about patient engagement in research. Involving consumers in all steps of research is of great interest to the research community, funders and policy makers and there is a need both to increase awareness, understanding and practice of involvement. This paper might contribute to this. The authors have conducted a “meta-narrative systematic review”. I am not familiar with this type of review and suggest the authors define the study type. They should also clarify whether the review includes a meta-narrative approach in the analysis of data, or whether the study type is a “meta-narrative systematic review”. It might be understandable that all research and other types of information within an area need to be mapped and disseminated, but a journal article publication might not be the best format. A systematic review is defined as a review that addresses a clear question and is carried out through systematic and transparent methods, including search, quality assessment and summarizing the evidence. Even though the authors refer and report according to some criteria from the PRISMA statement there is a strong need for improvement and clarification of the review process, as well as narrowing the research question.

Response:
As the reviewer suggested, we made clarifications in the title, abstract and methods sections to highlight that:
1) This is a systematic review (i.e., it followed a priori established protocol that defined inclusion criteria and outcomes that was developed by a) the study investigators, 2) patients and 3) experts from the Patient-Centered Outcomes Research Institute who developed the Request for Proposal (RFP) and funded the project).
2) The analysis was “meta-narrative synthesis” and not a quantitative one
3) Analysis plan is visually depicted in figure 1 that describes the 4 key questions of the systematic review. The methods section was heavily edited to explain the concept of meta-narrative reviews with key references.

We also re-organized the results section to reflect the 4 key questions in the same order. We added exemplars to every section to better illustrate inferences. We thank the reviewer for pointing out the need for clarity and we believe this improved the readability of the manuscript to a great extent.

Major Compulsory Revisions
1. The review question is extremely broad. It intends to summarize “evidence about patient engagement is research” and to include studies of any design. Comments, opinions, letters and editorials are also included as evidence, as well as reference to organizations and websites. These are publications without empirical data and would
normally not be classified as “evidence” or “studies”. The authors should consider excluding information without empirical data and clarify this both in methods and results.

Response:
We agree. In the revised manuscript, we excluded all non-original studies (non-systematic literature reviews, comments, opinions, letters and editorials, etc) as suggested by the reviewer.

2. The unclearness of the research question brings further confuse into both methods and results. One way to clarify might be to address the four questions identified within the analytic framework (Figure 1) into the aim of the review. Preferably I would like to see each question addressed in separate papers and with separate methods. Alternatively, if the authors still want to address them all, the questions must be addressed within the aim of the study. In the method section through the eligibility criteria each question needs to be outlined and specified for type of studies (design) to be included, assessed and summarized. Finally the result part should follow the same pattern by reporting findings according to each of the four questions and for each design.

Response:
We thank the reviewer for this suggestion. As depicted by the framework, we firmly believe that these questions are uniquely linked in the process of patient engagement. Separate papers or separate searches would cut the links between these questions and limit our ability to see the whole picture. Also we developed a broad search strategy (Appendix 1) to identify as many studies as possible. Separate searches on each question would lead to narrower searches and significant overlaps of the included studies between the questions. However, we agree with the reviewer and re-organized the results section to reflect the 4 key questions in the same order. We also added exemplars to every section to better illustrate inferences. The inclusion and exclusion criteria are spelled out in the methods section.

3. Since this topic already has been covered in published systematic reviews, an alternative solution for this paper could be to perform an overview of reviews only. Alternatively the authors could address the questions not summarized in published systematic reviews. This approach could also allow for assessment of risk of bias or other quality criteria depending on design. The rationale for not including risk of bias/quality assessment is not clear. This is an essential part of a systematic review.

Response:
Most of the identified systematic reviews (Appendix Table 2) were published before 2010. And all of them had restricted/narrower inclusion/exclusion criteria (settings, study countries, etc.) and couldn’t fully address our questions. We, as well as (PCORI) the study sponsor, determined that a review of systematic reviews couldn’t fully address our questions in this case.
Regarding the assessment of methodological quality of the included study, similar to our previous published systematic reviews, we had initially attempted to assess the methodological quality of the included studies using standardized scales (eg, the Cochrane Risk of Bias tool for RCTs, the Newcastle Ottawa scale for observational studies, etc); however, we quickly found that this assessment actually relates to the outcomes of the studies and not really to the outcome of patient engagement (the main purpose of this systematic review). Such assessment can actually be misleading. In other words, it is quite possible to have an excellent study (at low risk of bias for its primary and secondary outcomes) that has tokenistic patient engagement (i.e., not meaningful or helpful engagement). Future development of tools to assess the quality of patient engagement are needed. Such tools need to capture the outcomes of engagement, not just the process. These outcomes are not assessed in most studies at the present time.

We added this important point to the discussion section.

Minor Essential Revisions
4. According to the eligibility criteria this review looks at studies in which patients took part in design, conduct and dissemination of research. Patients should be involved in every step of the research circle. The important step of “prioritization” is not included or mentioned and should be addressed.

Response:
Actually many studies described involving patients in the prioritization of research topics and agenda. We included this under agenda setting. In the revised manuscript, we clarified these terms.

5. In Additional table 1 the included studies should be sorted according to their research question or their design rather than in alphabetical order.

Response:
We agree with the reviewer and sorted the studies by design in Appendix Table 2.

6. Please define “non-original” as publication type in the eligibility criteria part.

Response:
In our original manuscript, we define non-original study is anything other than original research. As we discussed above, we excluded non-original studies, except systematic reviews, in this revision.

7. Please clarify the aim of the environmental scan and Additional table 1. Was this scan carried out to identify more studies only, or to describe relevant organizations and sources for user involvement? Did the scan result in more studies included? If yes, how many studies?
Response:
The purpose of the environmental scan is to identify additional unpublished resources. In the revised manuscript, we clarified the purpose of the environmental scan and labelled the environmental scan table as “Initiatives and patient organizations identified by the environmental scan”.

8. The title could be revised and shortened into “Patient engagement in research..” leaving out “Patient Centered Outcome”. This phrase is not needed to address the topic.

Response:
We agree, title changed.

9. In the abstract the aim of the study is missing.

Response:
We added the aim to the abstract.
Reviewer's report
Reviewer: Alex Pollock

1. Analysis.
Figure 1 clearly defines the questions of interest and it is stated that a “meta-narrative approach” was used to analyse the data. Although a reference is used to support the use of this “meta-narrative” approach, the authors have failed to state or explain what they actually DID in order to analyse the results. What data was collected, by whom, and how was it categorised?? Was this done by one reviewer, two reviewers, by consensus? Was a particular method of analysis used – was the aim to aggregate or configure data? With the sort of data that the reviewers are synthesising I would have anticipated that they would have used some sort of thematic analysis to identify and collate the key themes and subthemes arising from the identified studies.

Response:
We thank the reviewer for this comment. We added a section, Analysis, in the Methods to clarify the analytical steps we adopted in this manuscript and highlighted key methodological references commonly cited for this approach.

2. Results.
This point relates to point 1, above. The questions of interest laid out in Figure 1 have not been answered comprehensively or systematically. Figure 3 summarises the types of engagement and the method with which this was explored; but there has been no attempt to explore the “best” methods, to compare methods, or explore issues such as the timing of engagement. While I appreciate that there were no comparative studies, I believe that you have synthesised substantial evidence relating to how different methods were implemented etc. The lack of systematic or comprehensive analysis of this evidence means that you have failed to adequately answer your questions of interest, despite having synthesised data which should be able to provide some answers if explored systematically.

Response:
The reviewer is essentially correct in that the available data fails to provide a convincingly superior approach. Perhaps this is because there is no one approach that fits all types of research. We do believe however, that it is very important to revisit this issue every few years and try to identify a good practice (probably not one, but several methods). In our revised manuscript, we added exemplars to demonstrate how engagement can be done, without pointing to a specific recommended superior approach.

3. Systematic reviews.
The authors state that they have identified 11 systematic reviews relating to public involvement in research. Each of these systematic reviews presumably summarises the results of a number of primary research studies, each answering a specific question
relating to public involvement. Unfortunately the authors have simply “lumped” these reviews in with the other studies, commentaries and opinion pieces. The authors have missed the opportunity to provide an overview of these reviews. Such an overview would provide an opportunity to assess the risk of bias within the individual reviews, as well as the quality of evidence included in the reviews.

Response:
We agree with the reviewer. We separated findings of the systematic reviews from the other studies and added a section in the Results to summarize these findings. For the issue of risk of bias, see next response.

4. Assessment of quality of included studies
The authors have not made any attempt to assess the quality of the studies that they have included. This is an essential component of any systematic review, and is crucial to enable appropriate interpretation of the results of the included studies. The authors state that “Studies reported design feature to assess the risk of bias for their primary and secondary outcomes; however, we found insufficient data to assess the risk of bias as it pertains to patient/informant engagement.” However there are several tools available for the quality assessment (critical appraisal) of studies, whatever their methodological design, and assessment of the quality of the included studies I believe is essential within any systematic review.

Response:
Regarding the assessment of methodological quality of the included study, similar to our previous published systematic reviews, we had initially attempted to assess the methodological quality of the included studies using standardized scales (eg, the Cochrane Risk of Bias tool for RCTs, the Newcastle Ottawa scale for observational studies, etc); however, we quickly found that this assessment actually relates to the outcomes of the studies and not really to the outcome of patient engagement (the main purpose of this systematic review). Such assessment can actually be misleading. In other words, it is quite possible to have an excellent study (at low risk of bias for its primary and secondary outcomes) that has tokenistic patient engagement (i.e., not meaningful or helpful engagement). Future development of tools to assess the quality of patient engagement are needed. Such tools need to capture the outcomes of engagement, not just the process. These outcomes are not assessed in most studies at the present time.
We added this important point to the discussion section.

5. Environmental scan.
I applaud the reviewers for attempting to identify the grey literature. However I am perplexed by the decision to report all evidence identified through this searching as separate from the electronic search results. The authors identified systematic reviews and papers within their ‘environmental scan’ – surely this evidence ought to be
synthesised and reviewed alongside the studies identified in the electronic searching? Just because it has been identified through a different route does not mean that it is of any lesser quality, or of any less relevance (and this would be formally assessed if you did a quality assessment as refered to in point 4 above). I appreciate that the websites and organisations are very clearly a different ‘type’ of evidence (perhaps more an information resource rather than ‘evidence’?) and agree that these ought to be synthesized separately from the research studies.

Response:
We agree with the reviewer. In the revised manuscript we moved published sources found through internet searching to the main tables and clearly separated the Websites, organizations, etc in their own table. This latter table is now titled “Initiatives and patient organizations identified by the environmental scan” for better clarity.

DISCRETIONARY COMMENTS
1. A lot of work has clearly gone into systematically identifying evidence and information sources relevant to public involvement. However in its current form I feel that the authors have not adequately addressed many of the essential components of a systematic review (i.e. assessment of study quality; systematic analysis of results). I feel that the authors have 2 choices:
   a) Revise the paper so that it is presented as a systematic identification of evidence and information sources relating to public involvement. i.e. the aim is simply to identify and list studies and information sources (which is what is currently done). This would meet the stated aim (page 4) of “synthesising the existing evidence about patient engagement in research” (but not the questions within Fig 1).
   b) Do additional work to appraise the quality of the evidence and systematically analyse results. In doing this I would recommend that the authors group their evidence according to the ‘type’ of evidence, which will enable them to select appropriate quality assessment tools and methods of analysis dependent on the type of study. This will help the authors reach informed conclusions based on the quality of the evidence that they have synthesised.

Response:
We believe that the reviewer will find the revised manuscript more satisfactory and clear. As mentioned earlier, this systematic review followed a priori established protocol approved by patients and by the funder (the Patient Centered Outcome Research Institute). Therefore, we are hesitant to make changes that reverse or undermine the intention of patients who reviewed and approved the protocol and identified the key questions. We agree with reviewer about the need for better clarity and we would like to point out the changes we did in the revision:
   1) Presenting results following the sequence of the 4 key questions
   2) Adding “analysis” section that describes the meta-narrative methods
3) Deleting non original and non-systematic reviews
4) Separating in the tables studies according to their design
5) Highlighting in the discussion section that we were unable to assess the methodological quality (risk of bias) that relates to the outcome of engagement (detailed response above)

We believe that these changes are important and thank the reviewers for pointing these out; we also believe that these changes did not significantly alter the manuscript in a way that is inconsistent with the desire of the patients involved in this project.

2. The authors have chosen to include studies where the informants’ role was as a participant. One could argue that these studies are investigating participation in research, rather than true “involvement”. I think that it would probably be impossible to identify all primary studies that have reported some degree of feedback from their participants, and I don’t believe that the search strategy will have comprehensively identified all studies where the experience of participation has been explored (due to inconsistencies in the reporting of this, rather than due to any failures of the search strategy). Since the authors’ questions are really about “engagement”, rather than “participation”, I do wonder whether one way to make this review both more manageable (in terms of its size) and more useable (in terms of the usefulness and accessibility of presented information) might be to exclude studies that are exploring participation, and concentrate on studies of user-engagement, collaboration and co-production?

Response:
We did not include studies of passive participation. We only included studies in which patients were engaged. The term “informant” has been used in the literature to describe a patient (or surrogate) who informs research. So, even in studies of active patient engagement, the term informant has been used. It is really not a good term for the very reason the reviewer brought up. To be more clear, in the revised manuscript, we deleted this term.