Author’s response to reviews

Title: Stakeholder involvement in systematic reviews: a protocol for a systematic review of methods, outcomes and effects

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Author’s response to reviews:

Reviewer #1: Page 8 line 147 comprehensive should comprehensiveness.

RESPONSE: Corrected

Reviewer #2: This is a clearly set out protocol for a study, which promises to produce useful information as part of a wider initiative for researchers conducting systematic reviews. Much of the language used, particularly in the long design section, is technical rather than Plain English, which will mean that it is unlikely to be very accessible to the patient and public readership of the journal but it is well laid out and can be followed by those with an understanding of the review methods described. My numbered comments below are all minor / discretionary:

1. At the end of the conclusions we are told the ways the results will be used but that isn't covered in either the Plain English Summary or abstract. It would be good if both said more than simply the results will "help researchers to improve, and evaluate, ways of involving stakeholders in systematic reviews".
RESPONSE: Added this in to both (plus minor edits to rest of these sections to keep word count within limits)

2. 1st sentence of both Plain English Summary and Abstract - I note the change made prior to peer review and understand what is being said but the sentence does not seem to me to be grammatically correct and is not easy to read. It is not clear who is the subject of the subordinate clause, researchers or those who expect them to involve stakeholders or both. I think it would be better to phrase the sentence differently or split into two sentences.

RESPONSE: Split into 2 sentences

3. The definition or systematic reviews (lines 109 and 110) doesn't actually explain what they are. The word 'synthesise' is jargon in this context and should perhaps be replaced with a description of what it actually means (preferably in Plain English). The purpose as set out in the second half of the sentence is clear but someone who doesn't already know what a systematic review is will be none the wiser after reading this sentence. The longer definition starting on line 216 is more informative but still does not define 'synthesise' in this context. Reference 26 does explain what a systematic review is more simply and in some detail and provides an explanation of what 'synthesise' means on another page (http://eppi.ioe.ac.uk/cms/Default.aspx?tabid=69), which could be included for those who don't know?

RESPONSE: The word synthesise has been replaced with “find and bring together”. In the longer definition we have put the word synthesised in brackets (after the phrase “brought together”) to provide greater clarity.

4. 'Synthesise' is also used in lines 125 and 130. As indicated above it would be helpful to define it for those who don't know what it means in this context (which will be most readers who are not researchers of whom there are a substantial number for this journal).

RESPONSE: We have changed these uses to “find and bring together” and “summarised”

5. I am not sure what the 'synthesise' in line 130 actually means either because the evidence to which it is referring has already been 'synthesised'. Is aim 2B to 'describe' the evidence on the impact of stakeholder involvement or perhaps use it in some way to produce a guide or toolkit to "help researchers to improve, and evaluate, ways of involving stakeholders in systematic reviews"?
RESPONSE: We have changed this to “summarised”

6. Line 156 - I too would expect to see a considerable increase in reports of involvement in systematic reviews over time in common with other areas of research but I doubt it would be ‘exponential’ even in other areas of research where involvement is more established.

RESPONSE: We have removed the word exponential.

7. Scoping review methods line 163, is the search really starting as recently as 2014? Or is that a typographical error? If it is right then why are you limiting it so much and not going back further into the history of systematic reviews? There are many examples of excellent practice in involvement going back a long time before 2014. Or is the search building on the one in reference 22 (which has now been published)?

RESPONSE: NO CHANGE – the first step of our search strategy is a 2-year period (2014-2016), but subsequent search steps expand the search in 2-year increments, going back in time.

8. Search methods - in addition to the list of established academic databases and other sources would it also be worth using Google / Google Scholar (and other internet search engines), especially for grey literature?

RESPONSE: NO CHANGE – we have given our search strategy considerable thought, and feel that expanding into internet search engines would be extremely challenging, and difficult to retain a focus on systematic reviews. Instead we have selected to contact authors, organisations and use social media to find grey literature, which we feel should be more successful.

9. Should the search methods also include a call for input from the readership of this journal, which I assume is one of the reasons for publishing the protocol for the study before undertaking it?

RESPONSE: NO CHANGE – we believe our statement about contacting experts etc should cover this. In the final line of our paper we do invite readers to contact us.

10. Definition of stakeholder, lines 205 et. seq. This repeats what is in lines 101 et. seq. but then expands on it. Maybe say ‘Further to the definition above…’ or start the first sentence ‘As stated above we define…’ so that the reason for repeating it is clearer?
11. Definition of involvement in a systematic review. I think that this section needs more thought because adopting a wide definition of involvement may make it more difficult to meet aim 2B to identify "evidence relating to the effect of stakeholder involvement on the quality, relevance or impact of systematic reviews".

RESPONSE: NO CHANGE – having this wide definition will enable us to create the broad map (aim 1). We will use this broad map to identify evidence relating to effect (aim 2B). We are confident that we will be able to identify evidence relating to effect from our broad map of evidence.

12. The problem may be that in including "reports relating to any role or contribution toward the development of a review protocol, completion of any of the stages of a systematic review, or dissemination of the findings of a review" the activities are likely to range from one-off consultations that could be meaningful or just as easily be tokenistic right through to extensive collaborative involvement throughout the process that is much more likely to have a positive impact. Further, this will also be dependent on how extensively the involvement is described. It is widely known within the public involvement community that descriptions of involvement in published papers vary widely, if they are included at all, and that the detail is not necessarily proportionate to the nature and effectiveness of what was done. That can make it very difficult to tell what was done, how well and what effect it had on the research. For that reason it would be advisable to document as carefully as possible the nature of what is reported as involvement so that some attempt can be made to categorise it with a view to being able to associate the impact on the systematic reviews. This issue is likely to make achieving the both parts of the second aim of the study quite challenging and is made harder by the lack of any agreed 'standards' for involvement, although there is work in progress in a number of organisations that might help within the timescale of the planned work.

RESPONSE: NO CHANGE – Thank you for this comment, which we are in agreement with. We have planned our ‘traffic light’ approach specifically in an attempt to deal with the issue of poor description of involvement. We hope that being able to report the volume of evidence which has a poor description will be useful for highlighting this issue further.

13. To an extent part of my concern may be mitigated by the categorisation of papers described in the section on data extraction, especially the first category of papers that focus on stakeholder involvement in systematic reviews. I would expect papers in that first category to
be the most useful. It is the other categories, which are likely to be the most numerous, that are likely to be subject to the wide variations I have outlined.

RESPONSE: NO CHANGE – thank you for this comment. Again we agree – and our intention is that the ‘amber’ and ‘red’ categories will contribute to the map, but not to other parts of the synthesis.

14. The criteria in Table 1 go some way to recognising the variability in what is reported but do not take account of the variability in the involvement itself and its likely impact on research. The information listed under data extraction will expose the variability of the involvement. It will be interesting to see how many papers actually report the level of detail sought, especially peer-reviewed papers. The grey literature may be more informative.

RESPONSE: NO CHANGE – we agree, it will be interesting to see both the variability in involvement and the level of reporting.

15. Also reference 22 seems to indicate that the feasibility of the methods and success in finding enough papers has been established? Perhaps this link should be made explicit? And how much did the scoping review in reference 22 address what this study is aiming to do?

RESPONSE: We have added a sentence into the Background section in order to clarify the difference between review 22 (now 14) and our proposed review.

16. Line 294, note that in the UK and for research conducted in the NHS approval is not required for involvement per se from an NHS Research Ethics Committee (REC) because their remit is to protect participants in studies. Any ethical issues related to involvement may be addressed when a study that needs ethical review is submitted for review. Research conducted outside the NHS is not within the remit of NHS RECs but will be within the remit of other ethics committees such as in universities. That does not mean that there are no ethical issues to consider.

RESPONSE: NO CHANGE. There is evidence of inconsistencies in ethical approval (and discussion around whether involvement is – in some circumstances – actually ‘research’). We therefore think it is appropriate and will be helpful collect data relating to this.

17. Abstract, line 80 - I don't think 'narratively' is a real word - couldn't find it in any dictionary I looked at. Perhaps replace with 'in narrative form' or 'as narrative'?
Reviewer #3: I thought this was a very interesting, well written and well set out protocol for a systematic review of stakeholder involvement in systematic reviews. I felt the authors have been very thorough in the development of the review questions and objectives, and in setting out their search strategy and plans for synthesis. Overall, I think the authors propose to conduct a study that will ultimately be of benefit in guiding and advising the systematic review community and will offer practical suggestions regarding how to improve stakeholder involvement in future systematic reviews.

I have only two specific comments / queries that the authors may consider / respond to, as follows:

1. Search methods (pages 8-10, lines 160-188)
I am interested in the stepwise approach to searching for eligible studies that the authors have presented. I think it seems very fair and is a very practical approach to performing very broad, comprehensive scoping searches. However, I would have liked to have seen some pre-defined decision rules in the protocol, e.g. for assessing redundancy of databases, or determining when further cycles of searching would offer minimal additional benefits. While the team decisions are being recoded for transparency, I wonder whether some more formal rules (akin to a stopping rule in a clinical trial) may be of use?

RESPONSE: NO CHANGE. We agree that more formal rules would be of use. However we lacked the evidence on which to develop such rules and therefore considered that a more pragmatic approach, with clear reporting of decision making, was the optimal approach. We hope to be able to report points of discussion which were used to make decisions, and that these may help inform the development of decision rules for use with this approach in the future.

2. Synthesis A methods (page 14, lines 274-277) It may be that I have not completely followed (or have misunderstood) the planned synthesis method, however the authors state that only studies judged green or amber will be included. If I have correctly understood Table 1 (Criteria for judgement of focus and comprehensiveness of reports of involvement in systematic reviews), those judged as 'Red' are studies lacking information on which to make the assessment. I would just query whether a better approach would be to obtain the missing information on 'red' studies (e.g. by contacting investigators) and making a final decision to include / exclude from Synthesis A based on all the relevant information obtained? I think this may be helpful, particularly if the authors retrieve fewer studies from their searches than they hope or anticipate.

RESPONSE: NO CHANGE. Unfortunately it will be beyond the scope of our review resources to obtain missing information on all the ‘red’ studies. Initial searches to suggest that we will retrieve sufficient numbers of green / amber studies to enable adequate descriptions of different types of involvement.

Reviewer #4: Pollock and colleagues have outlined a protocol for a scoping review of stakeholder involvement in systematic reviews, followed by a synthesis of methods of involvement and a synthesis of evaluation studies of involvement. The authors further outline that they will integrate the two syntheses by describing agreement and dissonance between studies that demonstrated a beneficial effect and had a comprehensive description of stakeholder involvement methods. This is a timely and useful study that aims to document existing practices
and impact and use this information to produce guidance on how to involve stakeholders in systematic reviews.

Specific comments

1) Regarding your consideration of databases: "Redundancy of individual databases will be considered in relation to the number of unique citations identified." It is understandable to limit hand searches to useful sources, but for databases why not run the searches and remove duplicates via reference management software?

RESPONSE: Thank you for highlighting this point, which was due to lack of clarity within our text. We have amended this sentence to clarify what we meant. It now reads “Redundancy of individual databases will be considered in relation to the number of unique citations judged to meet inclusion criteria”. We will be considering redundancy in relation to the number of included studies arising from each database, and not (as our sentence could previously be read) just in relation to the number of citations identified from the search.

2) In the authors' definition of a stakeholder (e.g. primary job is not related to research), it may be worthwhile to consider that someone who works in research should be able to comment about their lived experience as a carer, patient, or policy maker. This could be accomplished by stating that their primary job is not related to research, or their role in the systematic review is related to some non-employment role.

RESPONSE: NO CHANGE. We agree and we hope that our use of the phrase “primary job” will work to enable us to identify these people.

3) The authors mention that "Systematic reviews focused on synthesizing evidence relating to stakeholder involvement in primary research" are excluded, though it is unclear why primary studies exploring issues in stakeholder involvement in primary research would be eligible.

RESPONSE: NO CHANGE. We feel that this point is clear. We anticipate that our search strategy may identify systematic reviews which are synthesising evidence relating to stakeholder involvement in primary research. However this would not meet our inclusion criteria and would therefore be excluded.
4) For Synthesis B, what kind of outcomes are considered regarding the "quality," "relevance," and "impact" of the systematic review? It would be useful to include some examples.

RESPONSE: We have not changed the original text as we plan to be inclusive of any reported outcomes relating to effect or impact (based on author definition), and one of the outputs of our review will be a “list of unique methods for assessing the effect of involvement”. However we have added a sentence referring to a review by Brett 2014 which described the impact of involvement in research (more broadly), as we believe this provides a relevant example of how the results could be successfully synthesised.

5) Synthesis B seems to fit in to a standard assessment of interventions (this seems clear when the authors use GRADE to assess the certainty of the bodies of evidence). Given this is the case, Synthesis B should be framed accordingly. There should be a clear study question (or likely questions) with the "population" (this being, as we understand it, investigators conducting a systematic review), interventions (involvement of stakeholders in the review), comparator (no involvement of stakeholders) and the outcomes clearly specified. The authors should also state whether they contemplate any pooled analysis and if so how they will conduct it. Authors should make clear if they are going to consider involvement of any stakeholder as a similar intervention, or whether they would consider involvement of, for instance, patients versus front-line clinicians as different interventions.

RESPONSE: NO CHANGE. Although we are looking to include studies reporting “effect”, we do not anticipate that these will fit neatly into a “standard assessment of interventions”, and certainly we will be very surprised if we identify any prospective intervention studies. Rather we consider that we may find (for example) a questionnaire which has asked for opinions about effects. We have therefore deliberately framed this section in a way which will enable us to be inclusive of these sorts of studies. We feel that we have been clear where we say that we will document within a table any “outcome data” (quantitative or qualitative). We hope that framing this section in this way will give us a platform to highlight issues associated with the (lack of high quality) evidence (including lack of appropriate study designs) relating to effect of involvement.

6) With the advent of GRADE the term "quality" has become ambiguous. Traditionally, it has often been used for risk of bias in individual studies. GRADE has used it for the rating of trustworthiness of a body of evidence including considerations of risk of bias, precision, directness, consistency and publication bias. Because of this confusion GRADE has more recently used "certainty" rather than quality. It is probably advisable to use the term "risk of bias" for assessment of individual studies (we presume that is what authors are getting at) rather than quality. In the description of GRADE it should be clear that this is being used to
assess the entire body of evidence. The confusion here is particularly evidence in Figure 1 in which the heading "quality appraisal" refers to individual studies, and subsequently under outputs "quality of the evidence" is referred to. The latter, we presume is the GRADE assessment of the quality of evidence, which would be applied to the entire body of evidence for each intervention.

RESPONSE: Thank you for this comment. We have reconsidered our plan to assess the body of evidence using GRADE, and feel that this is probably overly ambitious given the sorts of evidence that we are anticipating and the challenges that we may face bringing varied ways of measuring outcome together. We have therefore removed all reference to GRADE.

7) Describing the studies that had a beneficial effect and comprehensive description of methods may be limiting in that those same factors could have not worked in other studies. Perhaps it may be worthwhile to consider focusing on commonly recommended methods for involvement, and reporting how many studies that used this method demonstrated beneficial effect, no effect, and negative effect.

RESPONSE: NO CHANGE. We feel that this is an issue which we should raise (if relevant) within the discussion of our completed review – i.e. whether or not any beneficial findings may be generalizable to other studies/settings/contexts. Within the discussion we would also plan to consider commonly recommended methods and the evidence which exists for these.