Author’s response to reviews

Title: Patient involvement in cardiovascular research: a qualitative impact evaluation

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Author’s response to reviews:

Dear Dr. Staniszewska and Mr. Stephens,

Please find enclosed the revised manuscript entitled “Patient involvement in cardiovascular research: a qualitative impact evaluation” to be submitted for publication in the Research Involvement and Engagement. We would like to thank the reviewers for their constructive feedback. We are delighted that they agree that the manuscript addresses an important and relevant issue. The reviewers made a number of comments on the use of English language. We regret these mistakes and apologize for any inconveniences this may have caused. We took the reviewers advice to carefully check the accuracy of the language used in the manuscript, and asked a professional editor for editing the English. Corrections of the editor are not incorporated in the tracked changes. The main suggestions of the reviewers were to provide clarification about the theoretical foundation of the study, strengthen and deepen the discussion on diversity, and training of the people involved, and elaborate on the limitations of the study. We have made changes in the manuscript, addressing the reviewers’ comments and questions. In the main document, these changes have been highlighted by using ‘track changes’. In order to maintain the readability of the manuscript, the discussion section was restructured. This restructuring is not incorporated in the tracked changes.

Below we provide a point-by-point explanation of how we have responded to the reviewers’ suggestions:

Authors’ response to reviewer 1

Reviewer one made a number of comments which we will address one by one:

1) Within the plain English summary: Point 3 'make patients advice more binding' I found hard to interpret what this meant - can the point be made more clearer in what you mean by 'binding' for readers?

The sentence ‘make patients advice more binding’ has been rewritten: “Making it obligatory for the researchers to clarify how the patient’s advice was incorporated”. This change was incorporated in the abstract as well.
2) Reviewer 1 addressed some questions on the definition and integration of the concept diversity in previous studies in PPI, and suggested to define 'process' from Brett et al in our own way and to include within it 'diversity', as the reviewer would have thought that ensuring diversity would fit under the umbrella of processes.

We have made some changes in the theoretical background to address these points:

“The process of patient involvement refers to its specific aspects of the methodology and execution, including the moment PPI takes place, by which design, the level of involvement, and the diversity of the people involved. Diversity is particularly relevant, as patients are not a homogeneous group. Different studies require different patient subgroups and different patient subgroups might require different involvement approaches and methods [14]. In this study, the aspect ‘level of involvement ’ was not interpreted as 'the higher the better', but as horizontal scale, due to the increasing criticism on the linearity of the participation ladder of Arnstein [15], which supposes higher levels to be better, instead of considering what is meaningful within a specific situation [16, 17].

The context of PPI refers to the conditions needed for involvement to work, including adequate funding and time, facilitating policy, adequate training and support and a positive attitude towards PPI [3, 6, 13].”

3) GRIPP 2 works well as an evaluation framework for impact of your work. Perhaps while you are responding to the previous point about diversity and process, you could amend the diagram 2 to include key things from GRIPP 2 for impact.

We recognize the previously narrow definition and operationalization of the concept impact. The GRIPP 2, as well and the GRIPP article from 2014 and the PIRICOM study, were helpful to clearly define impact, and highlight the focus we’ve had in our study: impact of PPI on de research design. We added a definition of the concept impact, and integrated this definition within figure 2:

“In previous studies on PPI, impact has been broadly defined as the influence or effect PPI has on aspects of research, researchers, organizations, patients and the public, service delivery and development, and/or evaluation [3, 13]. The current study primarily focussed on the impact of PPI on the research design.”

4) Could a reference be added to the Document analysis, Semi structured interviews and Focus group? This will show that the team are familiar with particular techniques in each of these methods.

We added a reference: Gray, 2014, as this book was used for designing and analysing both the semi structured interviews and the focus group. The two references which were already addressed within the method section, (Baarda 2009 and Hsieh 2005), are now addressed more frequently at the relevant sections.

5) The coding for the Figure two will need to reflect the point made earlier about the theoretical clarity asked for.

In line with comment 2 of the reviewer, on the aspect of diversity, figure 2 was adjusted as well as its description underneath.
Could the discussion section pick up the point about the panel members being highly educated and trained and the implications this might have on their layness and the discussion around diversity? In the UK there are debates about lay and scientific knowledge that patients bring and readers may find it helpful to know that you are mindful of such debates.

More information on the current training of members of the committee was added to the background section: “All members followed a two day training course given by a patient advocate, addressing the theory and practice of patient involvement, including how to critically read and comment on a research proposal from the patient perspective.” Members of the committee describe this training as helpful for providing advice, and simultaneously expressed the wish to regularly receive training on new developments within the field of research and participation.

Within the discussion section, both diversity and training became a separate paragraph, addressing the discussion on diversity and training more thoroughly. The authors elaborated on that the process of PPI ideally should accommodate all patients, from all groups of society. Moreover, we have elaborated on the different roles patients can fulfil within research, and that these different roles might require different knowledge and skills. PPI experts in the Netherlands are not univocal about if patients should receive training in the first place, and if so, how many training is ‘allowed’ to provide while maintaining the ‘pure patient perspective’.

Could you comment on how useful GRIPP 2 was for this study? For example reimbursements for patients involved?

We found the GRIPP2 useful when writing the article, to make sure all important aspects of the studied PPI were reported [line 146]. The information on how members of the committee are reimbursed was added in the background section.

Authors’ response to reviewer 2

Reviewer 2 made a number of comments which we will address one by one:

1) In terms of clarifications, I would suggest improving the introduction of section 3. I had a difficult time following this because of the language used in the introduction. I see that it is a continuation of the reporting of findings but is broken down by major theme / subject area, however this is not evident.

We have added a short introduction sentence to the sections of the results, to improve the readability of the results.

2) I would suggest including - in the discussion section - what implications this research could have for funding agencies (whether government or non-profit organizations) who are looking to engage patient partners as grant reviewers or patient partners in research. There is a lack of good information on what patient partners can bring to the table and your article highlights these areas well.

We thank the reviewer for addressing that our research could be helpful for funding agencies who are looking to engage with patients. In the discussion section, the following section was added: “The PPI approach of Harteraad is organised in an early phase of the research cycle, in the designing phase. It
would be of added value if patients have a role as co-assessor for grant requests as well, and we would like to recommend funding bodies to implement such an approach [26]. Similar to the current advice of the patient committee of Harteraad, patients would be able to assess the relevance, applicability and feasibility of the grant requests, alongside the scientific quality as assessed by a scientific committee. The Dutch Heart Foundation has already used such an approach of including the patient perspective in funding allocation.”

3) Also, potentially as part of the discussion, it may be helpful to identify the barriers to engage patients as part of this process. Were there any identified as part of this research? Example: volunteer nature, time commitment, compensation, the make up / composition of patient partners, etc.

Identifying barriers for patients to become member of the committee was not part of this study. However, we did observe/identify several barriers for participation. Not only in the level of education design of participation, but also in relation to severity of their disease, the energy available for participation and the amount of spare time a participant has available next to activities like a job, family care as well. Also the moment the meetings are organised influences the possibility to participate (e.g. during working hours or in the weekend). The observation of abovementioned aspects of diversity, and possible barriers to participate, are added to the manuscript [line 483 and further]

4) Although not necessarily important to the article itself, I really hope that there is the intention and funding dedicated to communicate the research findings to a broader audience than academic journals. Like I said, there are implications for funding agencies, NGO's, patient organizations - and not just in cardiology, but other disease areas - who would be highly interested in learning more about this project.

We thank the reviewer for this suggestion. The authors felt the need for communicating the results to a broader audience as well. Therefore we’ve written an article in 2017, in collaboration with PGOsupport, funding body ZonMw and Harteraad. This article aimed for direct improvement and implementation of directly formulated recommendations. The article is in Dutch, but if the reviewers are interested in reading it, it can be found here: https://participatiekompas.nl/wp-content/uploads/2017/11/17070_ZonMw-PP_brochureDEF_3.pdf

Authors’ response to reviewer 3
Reviewer 3 made a number of comments which we will address one by one:

1) Overall, I found the paper clear and fairly easy to follow. However, I would like to highlight some minor language issues:
   * Change 'subsidy' to 'funding' throughout.
   * Change 'The committee participates in implementing these recommendations' to 'committee members have contributed to implementing these recommendations' (abstract).
   * I'm not clear what the phrase 'to prevent a disguise of conflict of interest' means (line 154)
   * Line 171 - add the word 'for' after the word 'asking'.
   * Consider changing the term 'test-subjects' to 'research participants' throughout.
   * Consider changing 'Easier use of language' to 'use of simpler language' or 'use of clearer language' throughout.
We thank the reviewer for reading the manuscript thoroughly and addressing this language mistakes. All suggestions made above were incorporated in the manuscript. Moreover, a native speaker was asked for editing the manuscript.

2) The results section states: 'According to several interviewees, the input of patients should not be in the method and analysis of the research, since they consider that their own expertise' (line 246). However, I don't think that the quote which follows supports that interpretation. The researcher quote which follows that statement, seems to suggest that patient's views should be considered alongside scientific expertise; not that patients should be excluded from commenting on methods. I think that is a subtle but important difference.

We thank the reviewer for addressing this interpretation difference. Indeed the researcher of this quote considers patients’ input alongside scientific expertise. The interpretation of the findings has been slightly changed and is accompanied by another quote:

“According to several interviewees, the input of patients should not be primarily on the method and analysis of the research, since they consider that their own expertise. Researchers differ in their flexibility and willingness to consider patients’ advice on the method section: “I’d like to keep the choices for my research design and the analysis to myself.” (Researcher15), and “I was quite offended when patients commented on my power analysis [...] I know how to do a proper power analysis. (Researcher 14). Other researchers were more open for feedback on the methods and said to combine the patients’ input on methodological issues with the expertise of professionals and scientists. Most researchers suggested that the focus of the committee should be on the relevance, applicability and feasibility of the proposal and advice on distribution of the research results in the end.”

3) For me, a limitation of this study is that you conceptualise impact only in terms of impact on research, rather than impact on the people involved (researchers and public contributors). I think that limitation should be acknowledged.

The reviewers addresses an important point, which is in line with one of the comments of reviewer 1 on impact. The following lines were added within the discussion paragraph:

“Although our study mainly focused on the impact of patient involvement on the research (proposal), the findings also indicate that our approach has an impact on the researchers involved. For example, researchers increased their knowledge and experience on patient involvement and were stimulated to increase the involvement of patients during their project. Also, researchers were more motivated to involve patients again at the start of a new project. Impact on patients is often described in terms of empowerment, like acknowledgements of their experiences and opinions, they learn to critically reflect, gain confidence and increase their knowledge on medical topics [13, 14, 22] Although we recognise the impact involvement has on patients, this was not measured within this study.”

Moreover, we have recognized it as a limitation of our study: “Another limitation of this study is its scope on impact in terms of impact on research, rather than in the broader sense: on the patients and researchers involved. Although we did find aspects of the ‘intangible’ impact such as improved attitude for researchers, future research on the broader impact of PPI would be of added value.”

4) I was interested to read about participants' different perspectives on training. Training is a
contentious and complex issue within public involvement. I would like to see a little more critical reflection on training in the discussion (particularly pros and cons / approaches to training for committee members). I think you have started to explore issues related to this in your section on 'professionalization' but it could be linked more explicitly to training. This is important given that one of your recommendations is 'Improving training/education for researchers and the committee'.

We thank the reviewer for providing additional relevant literature to refer to in the discussion. The comment of this reviewer is in line with comment six of reviewer 1 on layness and training. As before mentioned, information on the content of the current training of the committee was added to the background section. This training primarily consist of information how the research process looks like, and how to use the feedback criteria the committee once formulated to use as a guidance. Improving training/education for both researchers and the committee is indeed one of the recommendations of this study. That relates to the currently used design of participation. In this design, members of the committee still feel the need for regular training on how to increase the quality of their advice and formulate it in such a way their recommendation and message is clear.

The discussion on training is extended and enriched with additional literature. The discussion starts at line 488.

Important to note is that we believe that when adapting the design of participation, less training is needed than with the current design. That is already covered in the discussion section.

5) It is not clear whether the study only included successful submissions or if those that incorporated lay reviewer's suggestions and comments were more successful.

The influence of PPI on the chances for funding allocation was beyond the scope of our study. In the method section the following sentence was added on the selection criteria of included proposals: “Whether or not the proposal of a researcher was honoured was no selection criterium.” This is also a limitation of our study, therefore the following sentence was assed to the discussion: “the relation between PPI and chances of funding allocation was beyond the scope of this study. As this would be interesting for both researchers, patients and the public, and funding bodies, future research on that relation would be recommended.”

6) The authors talk about research training needing to include modules on PPI. Training for researchers is not an homogenous route and there is no universally fool-proof way of ensuring all have PPI training. Do the authors have any suggestions of where that training might be optimally directed?

Based on some examples of training, we do have suggestions on how to organise training.

Therefore, the following section was added to the discussion: “In order to achieve intrinsic motivation of the researcher in the long run, patient involvement should become part of the academic curriculums of (at least) medicine, biomedical science and nursing. This will ensure that researchers are provided with the basics of patient involvement in their early career and are accustomed to the involvement of patients in their research [28, 30]. An example of how this could be organized is The Hague University of Applied Sciences, which integrated the course ‘participative healthcare’ in its curriculum for nurses. This course focusses on both the theory of patient involvement in healthcare, as well as the required practical skills. A similar course focussing on participation in research for(bio)medical students, could be an opportunity.”
7) We appreciate the authors are not writing in their own language and are very impressed with this. However, the writing lacks accuracy and/or clarity in a number of places and they need to rewrite carefully. For instance, the first sentence of the Abstract is missing a word: “Involving patients in scientific research has been shown to improve the relevance of the research…”

Later in the Abstract: “Researchers changed their proposals with i.e. the depth of patient involvement throughout the research…” Perhaps they mean “Researchers changed their proposals, for instance increasing the extent of patient involvement throughout the research,” but it is not clear currently.

We would like to thank the reviewer for noticing these unclarities. The suggested interpretation of the reviewer was accurate, and used in the abstract: “Researchers changed their proposals, for instance increasing the extent of patient involvement throughout the research.”

8) Final sentence of Abstract: “However, effort is required to add further weight on patient input and to support researchers in the required organizational and cultural change.” Again, meaning needs clarification: strengthen patient input, increased emphasis on responding positively to patient input, perhaps?

The sentence was rewritten to: “However, effort is required to increase the value that is currently attributed to patient involvement, to support researchers in the required organizational and cultural change and support them to meaningfully involve patients in their research.”

9) Please could the Background section end by clearly stating the aim of the paper.

The end of the background section has been slightly adapted, in such a way the research aim closes the background section.

10) Figure 3 should be in Results rather than Discussion.

We believe an analysis of the suitability of the used framework is a moment of reflection and therefore feel that this reflective moment should be in the discussion section. The clear finding of a relation between process and context is also a seen as a strength of our research, as it adds to the current framework. Hopefully the reviewer can agree with this line of reasoning.

11) The ‘strengths and limitations’ section needs to be expanded.

The strengths and limitation section has been extended. Two limitation were added: 1) the narrow interpretation of impact, 2) the fact that the relation between PPI and chances of funding allocation was beyond the scope of this study.
We think that our manuscript improved by these revisions and we hope that you will now find it suitable for publication.

We look forward to hearing from you,

Yours sincerely,

The authors