Author’s response to reviews

Title: Patient Involvement in Clinical Trials: Motivation and Expectations Differ between Patients and Researchers Involved in a Trial on Urinary Tract Infections

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Author’s response to reviews:

Dear editors,

Thank you very much for your feedback and the thoughtful comments of the reviewers. We addressed all of the comments. Please find our responses below each comment.

Kind regards,

Imke Schilling

REVIEWER #1

Dear Author/s,

First and foremost it was a privilege to review your manuscript. It is positive to see active research taking place surrounding this arena of work and in particular, strive to highlight necessity for joint partnerships to support best practice,

This a timely piece of work and believe that PPI is/ has become more important in the 5-7 years, (give or take a few years depending upon what type of research investigators are doing and where a team is in the world.
Given that your manuscript surrounded UTI's, I believe you should perhaps also relate some of the advantages/disadvantages of PPI and your key themes in the format of a summary table (will help readers to appreciate this more).

RESPONSE: We have added a table on key findings (table 3) in the results section.

You have made mention (which I think is quite pertinent): The external demand to conduct PPI (so that the criteria for donation or publication are met) might have an influence on the genuine interest of researchers in PPI. Further, when aware of this external demand on researchers, patients may get concerned that their involvement is a mere token involvement. To avoid misconceptions, researchers should reflect early on their motives and discuss them openly with patients.

Relating to the above, it would be good for the author/s to include some perspective on participation of patients being 'named co-author's on final publication', so that they have 'equal' credence, as much as the researchers.

RESPONSE: We agree with the reviewer, that co-authoring an article with patients is a very interesting issue and may be a good topic for further discussion. We were very lucky to have found two patients that were interested in getting involved as authors. They stated repeatedly that is was not the “fame” of being a co-author which got them interested, but their wish to help learning more about the topic. Inspired by your comment we added a further sentence to the methods section under 2.5 in which we describe how the patients became co-authors: “In addition, we asked all ten patients if some of them were interested in co-authoring this article. Two patients (HB and CH) expressed deeper interest to involve their perspectives in the preparation of the article to help with learning more about PPI. With them we intensively discussed the draft of the article. The patient co-authors contributed by giving valuable insights to the discussion of the results.” As working together with patient-co-authors is not the topic of our article, we did not elaborate on it any further.

You have also made mention (in your abstract):

Researchers were motivated by the possibility to improve research and to contribute to the empowerment of patients. They also wanted to enhance their career opportunities, to learn more about patient involvement and to meet the increasing demand for it.

Relating to the above, it would be good for the author/s to include some perspective on how PPI can support patient empowerment.

Considering researchers want patient involvement - then they also need to highlight how patients can support 'fellow patients' by getting involved in research-related activities.

For example, you have made mention of: It supports the empowerment of patients and leads to a democratization of research processes - how is the case, how are non-research patients encouraged/empowered?
RESPONSE: We read the suggested article with great interest and were deeply impressed by the work of the RPSG and its achievements for the community. However, we had the impression, that adding information on ideas to empower patients by the means of social media would go beyond the scope of our article.

If you address/ update your manuscript - it should be ready for publication.

Well done.

REVIEWER #2

It is a very good article on PPI and one can tell that patients were involved in the process. Particularly commendable is the involvement of patient co-authors. There is already a lot of similar research available in the UK, and the paper reflects all the common issues with PPI especially from the patients' point of view.

REVIEWER #3:

An interesting article which demonstrates good practice regarding patient involvement. Looking at both staff and patient perspectives simultaneously, then using the findings to facilitate the patient group may have increased the chance of effective PPI.

Need to re frame and change the title. It is not looking at patient and public involvement in clinical trials but at patient involvement in a UTI trial. This is true throughout the whole article. It is looking at patient Involvement not, patient and public involvement as there are no general members of the 'public' involved.

RESPONSE: We agree with the reviewer that the title might not be specific enough. We changed the title to ‘Patient Involvement in Clinical Trials: Motivation and Expectations Differ between Patients and Researchers Involved in a Trial on Urinary Tract Infections’. We deleted ‘public’ from the title. Even though we did not involve wider perspectives, e.g. potential patients or caregivers, we decided to stay with the term PPI in the further course of the article for reasons of consistency in the field of research. We added an explanation on the use of the term ‘PPI’ in the background section, where we already had defined the terms ‘patient’ and ‘researcher’: “Furthermore we used the term ‘patient and public involvement’ respectively its abbreviation PPI to describe the involvement of the ‘patients’ in the conduct of the UTI trial, even though we
are aware that the women that bring own experiences with UTIs into research are only a part of those who could get involved as the ‘public’. We did not involve further perspectives as potential patients or caregivers.”

The article is very long and while this is in part due to the fact that you are exploring both motivation and expectation from two perspectives, it does make it more difficult to read and to pull out your key findings. I suggest that you place some of the findings in tables and reduce the discussion.

RESPONSE: We agree with the reviewer that the article is quite long due to the fact that we report on both patients’ and researchers’ motivations and expectations. As we report the results of a qualitative study we found it hard to display the results in tables. Having this said, we added a table to highlight some key findings to the reader (Table 3: Expectations for involvement). This table displays the key findings of the section on expectations similar to the already existing table 2 that displays the key findings on motivation.

Furthermore, we significantly reduced the results section and shortened the discussion.

Lines 78, 81, 90, 91 and 93 are missing references. For example you state: "there have been few studies" but do not give any examples.

RESPONSE: We added references to the lines suggested. To enhance comprehensibility we rephrased the sentence in line 91 and added further information after the sentence in line 93.

Line 91: To date information on the motivations and expectations of patients and researchers when getting involved in PPI is mostly anecdotally described in studies reporting on PPI [4, 14, 17, 18].

Line 93: There have been few studies, which explicitly investigated motives and expectations for PPI: Tarpey reviewed articles on PPI and found personal and social reasons that influence the motivation of patients or the public to get involved in research [19]. Personal reasons refer to having a voice as well as the opportunity for personal development in terms of confidence, skills and self-esteem [19]. Social aspects reflected the wish to change research and services for the benefit of others [19]. Thompson et al. confirmed these results in a trial with patients and caregivers that were involved in cancer research [20]. Boaz et al. asked researchers about their attitude towards PPI [21]. They found that researchers’ attitudes ranged from the positive expectation that PPI improves research, to PPI being something that needs to be done to comply with a formal demand. While we found studies that described patients’ or researchers’ ideas of why to conduct PPI, we did not find any study that systematically investigated both patients’ and researchers’ motives and expectations.

Aims should be in methods section - as per the author instructions.
RESPONSE: Thank you for making us aware of this. To fulfil the author instructions we added the following sentence at the beginning of the methods section: “We aimed at studying the motivations and expectations patients and researchers in Germany have when establishing PPI.”

There is a line that mentions that the qualitative study is also part of a PhD project. Needs further explanation or removing.

RESPONSE: We agree with the reviewer that the information is misplaced. We removed the information and changed the sentence to: “Those who showed interest received information on the following aspects of the project in writing: the patient board and that it was a qualitative study.”

Twelve minutes are very short interviews to cover both topics (motivations and expectations). This is not reflected on in the limitations section of the report.

RESPONSE: The patients were free to talk as long as they wished in the interviews. The average time was twelve minutes. We agree with the reviewer that this is quite short. We assume that it is due to the fact that the whole process was unknown to the patients and it might be difficult to articulate specific expectations about something that is not known. It may be that the interviews would have taken longer, if we prompted them more, which we did not. We added the following sentence to the limitations section of the article: “The interviews with patients were short with twelve minutes mean to cover both their motivations and their expectations. Patients were free to talk as long as they wished. However, as the work in the patient board was unknown to them, it might be that it was difficult to articulate specific expectations.”

Theoretical positioning missing from methods.

RESPONSE: We have described our theoretical frame at the end of the background section: motivation is defined as a product of personal and situational factors. In chapter 2.3 we addressed the theoretical frame again when describing our interview guidelines. If the reviewer thinks we should refer to the frame somewhere else, we are happy to consider it.

Why telephone interviews not face to face?

RESPONSE: We conducted telephone interviews instead of face-to-face interviews for practical reasons. Some patients live out of town and some of the researchers live far away. We added an explanation to the methods section (2.3): “We conducted telephone interviews with each member of the patient board for practical reasons. Three out of ten patients lived out of town and four out of five researchers lived far away. Therefore telephone interviews were a more feasible method than face-to-face interviews. The semi-structured telephone interviews were conducted by the first author of this manuscript […]”
If you address these points with particular attention to the key finding and overall length, this paper will be a useful contribution to the PPI field.

REVIEWER #4:

This is a very interesting paper comparing researcher and patient motivations for involvement. I am not sure I agree that little is known about this topic. I was familiar with many of the issues raised but wondered if this is because of the conversations I have with researchers and patients/the public. Are the authors certain that there aren't any other published articles on this? How have they confirmed this?

RESPONSE: We agree with the researcher that there are already articles that report reasons for PPI from the perspectives of patients or researchers. However, we did not find any studies, which systematically investigated patients’ and researchers’ motivations and expectations for PPI. To add clarity, we rephrased the relevant section in the background of the article and added references for all sentences: “While the frequency with which PPI is being demanded and conducted is increasing [15], different papers report that even when people are willing to get involved in PPI, the effective realization of involvement can be hampered by differing ideas of PPI or the misinterpretation of aims and expectations among and between the parties involved [5, 16]. The values and ideas patients and researchers hold with regard to PPI influence the approaches used to conduct the involvement and the impact that results from it [5]. A mismatch can inhibit successful involvement [5]. To date information on the motivations and expectations of patients and researchers when getting involved in PPI is mostly anecdotally described in studies reporting on PPI [4, 14, 17, 18]. There have been few studies, which explicitly investigated motives and expectations for PPI: There have been few studies, which explicitly investigated motives and expectations for PPI: Tarpey reviewed articles on PPI and found personal and social reasons that influence the motivation of patients or the public to get involved in research [19]. Personal reasons refer to having a voice as well as the opportunity for personal development in terms of confidence, skills and self-esteem [19]. Social aspects reflected the wish to change research and services for the benefit of others [19]. Thompson et al. confirmed these results in a trial with patients and caregivers that were involved in cancer research [20]. Boaz et al. asked researchers about their attitude towards PPI [21]. They found that researchers’ attitudes ranged from the positive expectation that PPI enhances research, to PPI being something that needs to be done to comply with a formal demand. While we found studies that described patients’ or researchers’ ideas of why to conduct PPI, we did not find any study that systematically investigated both patients’ and researchers’ motives and expectations. The literature indicates that the view on PPI may vary quite strongly, from PPI being a value in itself, to PPI being a burden that needs to be carried [21-23]. To prevent conflicts, Gradinger et al. recommend that the parties involved in PPI explicitly reflect their positions and motives beforehand [5].”

Overall I thought the paper clear and well-written. There were a couple of places where the English wasn't quite the same as a native speaker e.g. it's the conduct of research, not the...
conduction of research. I would have added more comments on other examples, but was unable
to do that on the pdf version I had available. I suggest the paper is read again by someone whose
first language is English to make any other minor corrections. A note to the editors- is it possible
to access Word versions of the documents so as to be able to track changes of this kind?

RESPONSE: We asked an additional native speaker who identified the remaining mistakes,
which we corrected.

I also found the results and discussion sections a bit long and I got a bit lost in the detail. I
wondered if there was overlap between them or if they could be tightened somewhat. I suggest a
minor edit to try to do this.

RESPONSE: Following this commentary and the commentaries of reviewer 1 and 3 to display
the key findings in a table, we added a table to the result section of the article (Table 3: Expectations for involvement).

Furthermore, we significantly reduced the results section and shortened the discussion.

There were a few comments/ conclusions that I was unsure about. Firstly where the authors
referred to researchers not interacting with the panel, and someone not involved in the trial being
the communication link between the researchers and the panel. This is mentioned on page 23 and
page 27. In my opinion, if there isn't direct contact between researchers and patients/ the public
then the opportunities for mutual learning are severely restricted. This practice is more akin to
conducting qualitative research rather than involvement. Personally I wouldn't comment on this
as if this was Ok practice and I would want to highlight the limitations of doing this. If the
authors wanted to explore this, I suggest referring to the following articles:


RESPONSE: We agree with the reviewer that it is far from ideal that the coordination of the
patient board is with a researcher not involved in the trial. From the comment we furthermore
understood that we need to clarify our practice: One researcher attended all meetings of the
patient board and acted as a direct link between the research team and the patients on the board. All
other researchers were not obligated to attend each meeting of the patient board. We changed
the para on page 23 to “Some members of the trial team saw their own role as a “supporting
force” (T2:5), who does not necessarily participate in all patient board meetings but would get
informed about the results. Perhaps the fact that the coordination of the patient board was with a
person not involved in the trial had an impact on the way PPI was understood by researchers.
This may be different in situations where one of the trial coordinators also coordinates the PPI
for the trial. However, this form of conducting PPI with an independent researcher as coordinator
includes the risk that the benefit of PPI suffers from (too) little interaction between patients and
researchers. As Staley pointed out, the impact of involvement on research links to the impact the involvement has on the researchers: The interaction with patients allows researchers to learn and change their thinking [37]. This results in researchers doing research differently. Thus, if PPI does not allow for enough interaction between patients and researchers, it might not have that big an impact and might have more resemblance to qualitative research, in which data is collected [38].”

Furthermore, we deleted the sentence “It is important to establish structures for communication, especially for studies in which the PPI is conducted without a direct regular interaction between patients and researchers.” on page 27.

Finally on page 24, the authors report that researchers stated there was no need for involvement in the recruitment and data collection phases unless there was a problem. I'm not sure I'd agree - I think there is a need for ongoing discussions throughout all stages, as researchers cannot anticipate the problems they might face, nor be aware of their unknown unknowns ahead of time and therefore keep checking in with patients. Researchers should be asking themselves the following questions at all stages ‘have we missed anything? have we made any assumptions? are we on the right lines?’. The outcomes of involvement are not always predictable and therefore I'd conclude there shouldn't be any gaps in PPI, as researchers assumptions about when they need patient input might not be correct- again this issue is picked up in the above article if the authors wished to explore this.

RESPONSE: We agree with the reviewer that patients should be involved in all stages of the research process if possible. We added the following sentence: “It might be more advisable to involve patients at all stages of the research process and not only in the event of problems. Researchers cannot anticipate in advance what patients would add to the trial, they “[…] don’t know, what they don’t know” [37]”. The article suggested by the reviewer was of great use, thanks a lot for the recommendation.