Author’s response to reviews

Title: Establishing research priorities relating to the long-term impact of TIA and minor stroke through stakeholder-centred consensus

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Author’s response to reviews:

Dear Editors,

Thank you for your feedback. We have addressed the editor’s and reviewers’ constructive comments; details of revisions are listed below in a point-by-point format:

Reviewer #1:

I have used the Points to Consider in the Review Guidelines to structure my comments.

Is the rationale for what the author(s) have done clearly demonstrated?

Yes. I strongly support shared research priority setting and the implementation of it into the research cycle as commonplace for all the reasons set out by the author e.g. limiting waste research, transparency, relevance. The need for this in the area of TIA is as important as anywhere else. I think the UK could undertake shared priority setting a lot more than it does especially in light of Europe embracing Responsible Research and Innovation (RRI). I think this is a great example of how research priority setting can be done in a relatively time and (I assume) cost efficient manner compared to say a process like the James Lind Alliance Priority Setting Partnerships which take at least one year and cost approximately £30,000 which could possibly put researchers off involving all stakeholders in research priority setting.

-Thank you for these positive comments.
Have all methods been described in sufficient detail to allow others to evaluate and/or reproduce the work in similar circumstances?

It was not clear whether a systematic review had been undertaken in order to determine what research had already taken place in order to eliminate topics which had already been researched. Or was this done between the first and second stages after the stakeholders had submitted their list of priorities before the meeting?

-Prior to applying for funding for the stakeholder engagement event, a rapid review of the literature was undertaken. We found very few studies that addressed long-term impairments post-TIA and minor stroke. We have added a paragraph to the introduction to describe this (page 3 lines 22-32).

My other comments relate to the fact that the stakeholder group was limited to only 11 people (this limitation was noted in the submission although no reason given as to why it was this size). I would also question the number of each type of stakeholder involved. There were only 3 patients compared to 7 or 8 professionals (depending on whether the Stroke Association representative could be considered to be a patient representative or not).

-The sample size for the stakeholder event was restricted due to time constraints which limited the number of number of people we were able to recruit in a short timeframe. We have added an explanation of why the sample size was small in the limitations (page 6, line 21). Although there were only 3 patients they still had the second highest representation from the stakeholder groups (2 nurses, 2 doctors, 1 researcher, 1 psychologist, 1 Stroke Association representative).

I would have expected that carers would have been included in this stakeholder group and that the number of carers and patients would have at least equalled the number of professionals. One reason for this would be so that patients do not feel outnumbered by professionals resulting in lack of confidence to contribute fully or defend their opinions in the face of differing professional opinions.

TIA and minor stroke patients functionally recover well and the long-term effects of are often subtle or ‘hidden’. Therefore, it is not common for ----TIA/minor stroke patients to have carers. Although there were only 3 patients, we ensured that there was one patient per subgroup to ensure patient representation in the interim prioritisation. Furthermore, the subgroups were facilitated by researchers to ensure that everyone had an equal voice.

It is not stated what the make up of stakeholders was in each sub-group in the second stage. I think these details are important to know for the purposes of understanding possible power dynamics within each group and whether these played any part in which priorities were ranked in what order.

-We have clarified the make up of the subgroups on page 5 (lines 10-12). We have also added a sentence to describe how having a patient representative in each subgroup and a dedicated facilitator ensured that everyone had an equal voice (page 6, lines 18-19).
Is it clear exactly what was done, at what stage and what the outcome was? If anything is not clear, please provide feedback as to what needs clarifying.

See my response to the question above.

-See above

Has sufficient attention been given to ethical considerations and how these were managed?

In light of the fact that patients who have lived through TIA experience memory loss and fatigue, it was curious that this research priority setting exercise was conducted in one day instead of over a longer period e.g. 2 or 3 half days to mitigate any fatigue of the patients involved and hence maximise the quality of their responses.

- The advantage of a one day event is to maintain the flow of debates and discussions, reduce the time and resources required and reduce the burden on participants attending. Multiple breaks and refreshments were scheduled and all participants were told at the start of the event that they were welcome to take breaks at any point. We have added a sentence with a rationale of the one-day event to the methods (page 4, lines 7-8).

Can the writing, organization, tables or images be improved?

I do not have any comments in relation to this.

Are the included additional files (supplementary materials) appropriate I do not have any comments in relation to this.

Reviewer #2:

This is a worthwhile piece of work which I think could usefully inform the TIA/minor stroke research agenda, since it appears to be the first research prioritisation exercise in this area. The paper has a clear structure and is well-written. I do have some suggestions for improvement (listed below), particularly regarding the clarification/detailing of methods. I also think that this is an interesting paper methodologically, because it presents a relatively quick and cheap way of prioritising research questions. It would be helpful if the authors could reflect on the pros and cons of this approach, any learning points and implications for future research priority setting exercises.

Abstract

1. '…the three subgroups' - perhaps say what/who these were, or drop the word 'the', as the subgroups haven't been mentioned before?

-Reference to the subgroups have been added in the methods paragraph (page 2, line 16).
2. 'These research questions should be used to inform the research agenda…' As there are some important limitations, I wonder if this could be worded slightly more tentatively… perhaps 'could usefully inform the research agenda'?

-Thank you for this suggestion, the wording has been updated (page 2, lone 31).

3. I'm not sure I would go as far as saying they should inform policy decisions… What kind of policy decisions? Can this be justified?

-We would argue that the research priorities are very relevant to health policy, particularly regarding provision of a follow-up pathway for TIA and minor stroke patients.

Plain English summary

4. I think it would be helpful to mention what the prioritised research questions were in the 'What did we find?' section.

-A paragraph has been added to summarising the research priorities (page 1, lines 24-29).

Background

5. '…an episode of neurological dysfunction with evidence of acute infarction' - As this is a non-medical journal, perhaps this could be translated into more 'lay' language?

-Thank you for this comment, the wording has been amended (page 3 lines 4-5).

Methods

6. Why did you decide to group TIA and minor stroke together rather than focusing on one or the other? Is it possible that research priorities could differ between the two conditions?

-The two diagnoses were included together because both patient groups will experience similar ongoing residual impairments (as demonstrated by the literature (page 3, lines 16-21), it is the underlying biological mechanism which is different. Many experts argue that the two should be considered a continuum not distinct diagnoses. A sentence has been added to clarify this on page 3, line 7.

7. Why did you choose a one-day meeting / Nominal Group Technique rather than a more rigorous process such as a James Lind Alliance priority setting partnership? There may be good, practical reasons but I think they should be stated.

-A James Lind Alliance priority setting partnership is very costly and time consuming, we had neither the time nor resources to do this; therefore, a one day event was selected for pragmatic reasons. The rationale for this have been added to the methods (page 4, lines 7-8).
8. Please describe how the participants were recruited/selected. In addition, was there any prior or existing relationship between the patients and healthcare professionals which could have affected the balance of power? And what about existing relationships between the professionals in the group which could have led to 'group think'? Please reflect on these aspects as limitations if relevant.

-Recruitment strategies have been added: page 4, lines 9-13. There was no prior/ existing relationship between the patients and healthcare professionals. The stroke nurses knew each other; however, they were separated over two subgroups and the experienced researchers who facilitated the subgroups discussions ensured that everyone had an equal voice.

9. Gathering research priorities - Were any suggestions filtered out at this stage i.e. did not make it to the interim prioritisation stage? If so, what criteria were used?

-Only duplicates were removed at this stage, clarification of this has been added to the methods (page 4, line 24).

10. What was the composition of the subgroups? Did you group people according to their role? How was consensus on the order of priorities reached within subgroups?

-The composition of the subgroups have been added (page 5, lines 6-10). The consensus on order of priorities was reach through group discussion.

11. 'As a whole group, a final consensus on the shared priority areas was agreed.' - Please describe the process by which consensus was reached, and your definition of 'consensus'.

-In the Plain English summary you mention voting.

-The wording in the lay summary has been changed as this language is misleading as there was not a formal vote. The consensus was reached through group discussion with all 11 members and through this discussion there was mutual agreement on which research priorities should be included in the final list.

12. How did you ensure that the more dominant individuals did not have undue influence, or could this be a limitation?

-The subgroups and whole group discussions were facilitated by experienced researchers who ensured that all participants had an equal voice (detailed on page 4, lines 29 and 33).

13. The discussions were audio-recorded and I wonder what these recordings were used for? Were they transcribed and analysed at all? They could provide some useful information, such as how much 'air time' each individual or stakeholder group had (was this roughly equal or were there large differences?) and how decisions were arrived at.
- The discussions were recorded to document the event. The recordings were transcribed but have not been analysed. Analysis of the transcripts will be considered for a future research project, but is outside the remit of this paper.

Results

14. It would help to know which region(s) the participants came from. Was this a local or national group? If local, this should be mentioned as a limitation, since different regions may have generated different priorities due to differing healthcare services.

- The majority of stakeholders were from the West Midlands, this has been included as a limitation (page 6, lines 21-23).

15. Table 1 - Instead of having an 'other' stakeholder category with N=2, could you write out what each of individual's role was?

The table has been updated.

16. Table 2 - Are these questions ranked in order of priority? If so, please describe in your Methods how the ranking was agreed. If not, perhaps use bullet points instead of numbering to avoid confusion?

- The research priorities are not ranked in order of priority, the numbering has been changed to bullet points as suggested for clarity (see Table 2).

17. It would be interesting and useful to see which stakeholder group(s) each of the 11 priorities originated from (e.g. patient, healthcare professional, researcher etc.) - would it be possible to show this?

- Unfortunately this was not documented.

Discussion

18. 'The top agreed research questions provide valuable insight into the priorities of patients and key stakeholders…' As there were only 3 patients out of 11 stakeholders, I would avoid emphasising this group in particular. I think '…valuable insight into the priorities of key stakeholders' would be more appropriate.

- This has been updated (page 6, line 1).

19. '…the majority which were patients and healthcare professionals' should be 'patients or healthcare professionals'?

- This has been updated (page 6, line 15-16).

20. '…with limited social acceptability bias' - How do you know this?
The methodology used was specially chosen to reduce social acceptability bias, including individual ranking, mixed stakeholder subgroups and presence of experienced researchers as facilitators.

21. I agree that the small sample size of stakeholders is an important limitation. I would add that this means some important priorities may have been missed and that we cannot be sure the findings would be replicated in a different group. I would also add (if you agree) that only 3 patients were involved and ideally they would have constituted a larger proportion of the group. The low proportion of patients (and possible power imbalance) could explain why many of the resulting priorities have a distinctly medical focus.

Although there were only 3 patients they still had the second highest representation from the stakeholder groups (2 nurses, 2 doctors, 1 researcher, 1 psychologist, 1 Stroke Association representative). Furthermore, there was a patient representative in each subgroup. Arguably the majority of the research priorities are patient focused rather than medically focused, including: patient follow-up, identifying people with impairments, treating people with impairments, support groups, information giving after diagnosis, impact on families and impact of diagnosis on patients’ care.

22. It would be helpful if you could also reflect on the advantages and disadvantages of the approach you took as compared to the more time- and resource-intensive James Lind Alliance priority setting process.

We have added few sentences to discuss this on page 6, lines 23-29.

23. What did you learn from this exercise and what recommendations would you give others wishing to replicate this process for a different medical condition?

This was an extremely useful exercise which was beneficial not only to identify research priorities, but also to develop collaborations with different stakeholder groups. Have added a paragraph which discusses recommendations (page 6, lines 30-40).

Conclusions

24. This is currently a summary of the whole paper; I would include the a summary of key findings and potential implications only.

The conclusion has been updated to accommodate your suggestion.

Thank you and good luck!

Thank you

Reviewer #3:
This is a very clearly written paper on an important topic which I agree we have little understanding of. To undertake an exercise such as nominal group technique is an appropriate way to address the need to better understand the research priorities of this population. I think it is close to being of publishable quality. Its main weakness as you say is the small number of participants and it would have been much stronger with more patient representatives to gain more of their perspectives and to minimize risk of the discussion being dominated by the 8 non patients around the table. However we do these small scale exercises and they can be valuable, especially locally and to others who do not need (or have the funds for) a rigorous approach such as that by the James Lind Alliance to inform their research plans. How were the participants selected?

-Recruitment strategies have been added: page 4, lines 9-13.

What I would like to see is more depth about the issues participants said needed researching to bring it to life. For example, what is the problem with 'information' that requires research? What impact did they say TIA has on family members for that to warrant further research? Why do they think professionals need training? This would then move it on from being a report on an event to be a useful discussion of the issues raised.

-We agree that this information would be of interest; however, it would require analysis of the recorded subgroup discussions which is outside the remit of the current paper. Analysis of the audio recordings will be considered for a future research project.

It sounds like you had a great event and I can see much utility in the research questions you have come up with. I think readers would like more insight into what was discussed is all, ie the nature of the problems that need researching.

Reviewer #4:

This is an interesting and well written study.

As pointed out by the authors themselves, the main study limitation is the small sample size (11 stakeholders). Why was the sample size not larger?

-The sample size for the stakeholder event was restricted due to time constraints which limited the number of number of people we were able to recruit in a short timeframe. We have added an explanation of why the sample size was small in the limitations (page 6, line 21).

In particular, why were there no minor stroke/ TIA carers specifically included in the consensus workshop?

-TIA and minor stroke patients functionally recover well and the long-term effects of are often subtle or ‘hidden’. Therefore, it is not common for TIA/minor stroke patients to have carers.
As patients were not recruited through the NHS, I recognise that NHS Health Research Authority ethics was not required for this study. However, I am unclear as to why the university ethics was not obtained through a health related department or faculty.

-This is standard practice for research conducted at a University.

Did the researchers consider conducting their study in partnership with the James Lind Alliance. Why was this not done?

-We did look into the James Lind Alliance; however, their process is very costly (>£40,000) and time consuming (12 to 18 months). We had neither the time nor resources to partner with James Lind.