Reviewer’s report

Title: Rethinking Strategies for Positive Newborn Screening Result (NBS+) Delivery (ReSPoND): a process evaluation of co-designing interventions to minimise impact on parental emotional well-being and stress

Version: 0 Date: 06 Feb 2019

Reviewer: Jessica Jarvis

Reviewer's report:

This manuscript is the protocol for a study aimed at improving the way in which results from Newborn blood spot screening are conveyed to parents. The suggested approach involves the engagement of both parents and various medical staff through multiple phases of study design to develop an appropriate method for clinical uptake. In addition to the clinical significance of this work, the methods stand as a good example for the development of other clinical interventions. As written, however, this protocol requires significant additionally for clarity on methods and editing for clarity in reading, as well as edits for brevity prior to publication (the background is too long). Additionally, there are some enrollment procedures that may need revision in order to improve the generalizability of this intervention. Lastly, the data analysis plans in some phases require further detail (e.g., 'descriptive statistics' and 'content analysis' are too broad to provide meaningful insight). Thank you for the opportunity to review this manuscript. I have provided the following comments as suggestions for clarity and followed by questions for methods.

CLARITY:

Please read through entire manuscript with a careful eye with editing (e.g., titles of figures and tables are inconsistently capitalized, significant number of inconsistent reference styles throughout the entire manuscript, and random typos, e.g., line 355)

The manuscript is very lengthy and care should be taken to be as succinct as possible. For example, the background is thorough, but far too lengthy at approximately 8 pages. It should be much briefer, only as long as needed to provide a summary of why this is important, what's been done, and what gaps this study is addressing - approximately 2 pages. This would provide more space to elaborate in the methods.

I recommend removing the headings of "aims" and simply closing the background with a few succinct sentences on this study's aims. I recommend removing the objectives and proposed
outcomes subheading and content from the background all together. This can be elaborated on in the methods.

Similarly, in methods, I would remove the subheading of Aim and its content, as it is just written above.

To keep the methods from being unnecessarily length, I would start by listing what is consistent between all phases and not repeating under each phase (e.g., consent will be gathered for all participants at each phase, this can be simply stated prior to phase descriptions).

Please reorganize methods content for clarity. For example, under the subheading of 'setting' there is information on study sites, but also on condition groups, and study design. This makes reading through the content confusing.

Under Phase 1 subheading participants, the description is confusing, suggest rewording for clarity. Furthermore, the description of how participants are recruited could be combined with this section and would assist with clarity when reading.

Some acronyms are used without spelling them out first (e.g., PIS PPI, MRC. For clarity of content, I suggest not creating an acronym for a word that is used less than four or five times (e.g., PPIAG and PAG are spelled out as an acronym in line 667 and then used as an acronym only one additional time in text). To that end, not all of the acronyms used are listed in the abbreviations.

Suggest removing table two and using a few sentences to write out inclusion and exclusion criteria. Tables are helpful if they are a summarizing content in a manner that is easier and more efficient for comprehending than a written sentence. Table two is just as lengthy as a written sentence and is more difficult to read in this format.

Suggest removing the description in the first row of table 3 and simply writing the steps of the modified NGT process. Also, please write out NGT in the title

METHODS:

For all the phases, it is clear that there will be deliberate recruitment across the CSG's, but in order for the results and the intervention to be generalizable to the public as a whole there should be a clear description of attempts to recruit a diverse sample (by age, gender, ethnicity, socioeconomic status, etc.) It is unclear what demographic data is gathered, I would suggest gathering information on their experience in medical field (is the mother a nurse or someone who has never been in the hospital before? Is this their first child? Do they have other children with medical needs at home?)
Phase 1:

- Please provide further detail to the analytic plan. What will you be doing for "descriptive analyses" and will your content analysis be inductive or deductive? If inductive, what will be your unit of analysis/how will you create categories? If deductive, provide details on the creation of your matrix.

- It is unclear how these analyses will provide information on the "total cost of existing communication strategies" for use in phase 3 or how these costs will be compared (line 655).

- Phase 1 output says this phase will help you select relevant study sites - how will these study sites be determined? This should be clearly outline in the data collected and analysis process.

Phase 2:

- It is unclear why you are sampling 20 staff and only 15 will be interviewed/invited to the parent event. Additionally, what is meant by "non-participant observation"?

- Line 423 says that 20 parents who have been interviewed will be invited to the parent event. However, there is no description of parent recruitment in either Phase 1 or Phase 2.

- Why won't the parent videos have the faces pixelated to protect privacy?

Phase 3:

- What analytic approach will be used for economic evaluation?

- Line 532, please describe "pro forma"

- The other phases had a specific N, why is phase 3 a range of 20-30 and 20-25?

- Lines 270 and 581 mention potential measures, while I recognize these may not be the selected measures for a future definition trial of the created intervention, the measures that will be compared with the results of the qualitative analysis should be specifically stated and described.
- Lines 585 - 594 the process is confusing and unclear for me. If the content analyses us deductive, what matrix are you using?

- How you are determining feasibility and acceptability (output ii) is not clear in figure 3. E.g., figure asks if the training "requires too much time", what is the cut off for 'too much'? What determines if this is acceptable? And acceptable to whom, staff or parents, or both?

Phase 4:


- Line 619, do you mean 10 staff and 10 parents, or 10 people total?

DISCUSSION:

- Thank you for your description of ethical issues. I think it is a strength that you will have a clinical psychologist consultant in the case of a parent becoming distressed.

- Am I understanding lines 666-670 correctly that this will be a longitudinal study with follow-ups every 6 months? Every 6 months for how many months? How will "success" of the project be defined? Will parents continue to be involved during these follow-ups?

- When discussing potential impact and outcomes, this should pertain to the clinical relevancy of the results and not the fact that results will be disseminated (also, while I don't suggest listing dissemination as an outcome, if stated it should be written as a plan and not a certainty - you do not know for sure that this study will be accepted and published by the listed journals).
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Please indicate how interesting you found the manuscript:

An article whose findings are important to those with closely related research interests

**Quality of written English**
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Acceptable

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