Author’s response to reviews

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

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Author’s response to reviews:

Dear Professor Veroniki,

Thank you for the opportunity to resubmit a revised protocol for our study; 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) that we would like considered for publication in BMC Pilot and Feasibility Studies. This study is funded by NIHR HS&DR https://www.journalslibrary.nihr.ac.uk/programmes/hsdr/165225/#/.

I confirm the revised work has been seen and approved by all co-authors. As the changes are quite substantial, please let me know if you would like me to submit a 'clean' version with no track changes for clarity. Please see a point by point response below:
Reviewer #1: Thank you for the opportunity to review this well-written and interesting manuscript.

I believe the proposed study deals with an exciting topic and the findings of this study are likely to provide an important contribution to the field.

The intervention development is based on suitable theoretical frameworks and models, both parents and health care professionals are involved in the process. The methods which are proposed for intervention development and process evaluation are clearly described.

Thank you for your feedback.

Reviewer #2: 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 additional 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.

Thank you so much for your detailed feedback, it has been most helpful.

CLARITY:

1. 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)

A very careful proof read has been undertaken and inconsistencies have been corrected. Apologies for this oversight.
2. 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.

The background has been reduced substantially to approximately 2 pages as suggested.

3. 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.

The heading “aims” has been removed and the background has been closed with a sentence detailing the aim of the study. The objectives and proposed outcomes subheading and content from the background have been removed.

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

The subheading of “Aim” and its content have been removed from the methods section.

5. 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).

The following subsections which contain information which is consistent between all study sections have been added towards the beginning of the methods section: setting, inclusion and exclusion criteria, recruitment, demographic data and consent.

6. 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.

This has been reorganised and your helpful suggestion above (5.) has been utilised to improve clarity.
7. 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.

This has been reworded to improve clarity. A section on recruitment has been added at the beginning of the methods section to improve clarity and reduce length as suggested above (5.).

8. 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.

All acronyms have now been spelled out the first time they have been used. In addition, any acronyms that have been used less than four times have been removed. All of the acronyms that have been used, now appear in the list of abbreviations.

9. 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.

Table 2 has been removed and the content has been moved to a subsection at the beginning of the methods section as per your suggestion above (5.).

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.

The first row in Table 3 (now Table 2 has been removed) and NGT has been written out in full.

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?)

Information regarding the collection of demographic data has been added at the beginning of the methods chapter as per your comment above (5.).

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.

Further details have been provided about the descriptive analysis for Phase 1. The content analysis will be inductive and the process of code and category development has been described.

- 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)

To clarify this, the following information has been added to data analysis in Phase 1: “…[the] total cost of existing communication strategies, from the NHS perspective [will be calculated] by determining the grade of the person involved in the communication, the time taken and resources used.” The following information has been added to the discussion “This will include comparisons between costs of different approaches currently used (from Phase 1) and costs of the new, co-designed interventions in terms of grade of staff involved, time taken and resources used (Phase 3).

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

To clarify this, the following information has been added to data analysis in Phase 1, “These data will be presented to members of the study steering committee and the lay advisory group who will be involved in the decision regarding which study sites will be used in subsequent study Phases.”

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"?
This has been altered so that 15 staff will be observed, interviewed and invited to the parent event for consistency and in line with previous successful EBCD projects. The use of non-participant observation has been clarified in the Phase 2: Co-design section which now reads, “When the relevant member of the clinical team contacts the family to communicate the initial NBS+ result (by whichever methods they normally use e.g. phone or face-to-face), they will ask the family at the beginning of the interaction, whether a member of the research team may be present. If the family agree, a researcher will observe the clinician communicating the NBS+ result to the family. During the communication, the researcher will not participate in the interaction between the clinician and the family but will take detailed field notes.”

- 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?

A section on recruitment (staff and parents) has been added at the beginning of the methods section to improve clarity and reduce length as suggested above (5.).

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

Previous EBCD projects have not pixelated the faces of participants who have been interviewed. This allows staff to observe both verbal and non-verbal cues when they watch the interviews during the joint staff-parent event. It also allows the videos to be more personal and real in terms of it not just being a voice that is being listened to but a real person sitting in front of you.

Phase 3:

- What analytic approach will be used for economic evaluation?

We have added the following to describe the analytical approach, “Our analytical approach will be to undertake a cost analysis of the intervention coupled with a feasibility study to plan the economic evaluation that would accompany a definitive evaluation study. The analytical approach that we envisage using in the definitive study would either be a cost-utility analysis or a cost-consequences analysis, and which of these will be most appropriate will be determined during the present feasibility study.”

- Line 532, please describe "pro forma"

The pro forma has been described as an ,“… electronic document that upon completion, can be emailed directly to the study team.”

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

The range has been removed.
- 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.

The wording has been changed in the data analysis section for Phase 3 so now it is clear that these are the measures that will be compared with the results of the qualitative analysis. Each scale has also been briefly described.

- Lines 585 - 594 the process is confusing and unclear for me. If the content analyses us deductive, what matrix are you using?

This should have read inductive not deductive and has been changed accordingly.

- 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?

The wording in Figure 3 has been changed so that the question is now, “How much time is required for training on the new interventions and is this feasible in practise?”

Phase 4:


I agree. It was written this way following previous feedback from the funders which stated that the need for an evaluation study would not be known until at least some data had been collected. This has now been changed back so it reads, “…the nominal group technique (NGT) [will be] used to inform the design, of an evaluation study of the co-designed interventions.

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

This has been changed so it now states (n=10) to provide clarity.
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.

Thank you.

- 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?

It is not longitudinal, this has been clarified by stating that it will be “…for the duration of the study.”

- 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).

The dissemination plans are now written as being intended rather than being a certainty and have been moved so they appear before the discussion rather than being perceived as a proposed outcome.

The authors declare no conflicts of interest.

The study sponsor had no involvement in the, 1) study design; 2) collection, analysis, and interpretation of data; 3) writing of the report; and 4) decision to submit the manuscript for publication.

Dr Jane Chudleigh wrote the first draft of the manuscript and attended to the reviewers comments.

If you have any further questions, please do not hesitate to contact me.

Yours sincerely,
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