Author's response to reviews

Title: Imparting carrier status results detected by universal newborn screening for sickle cell and cystic fibrosis in England: a qualitative study of current practice and policy challenges

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Author's response to reviews: see over
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Dear Editor,

We are happy to respond to the points raised by yourself and the four reviewers and have made changes accordingly. Comments have been addressed as follows:

**Ethics and consent**
This information is now included in the ‘methods’ section of the paper as requested and type of consent specified

**Referee 1: Bridget Wilcken**

1) **Background paragraph 2:**
This paragraph has now been changed to clarify the CF screening protocol and that it refers to English practice.

2) **p5 A role for non-specialists:** The footnote is now included in the body of the text

3) **Revision of format of verbatim quotes:** We have not changed this as our preferred approach when reporting verbatim data in qualitative research is to include everything said, including pauses. This ensures authenticity of the quote. Paraphrasing verbatim data, although seemingly an issue of practicality, has the potential to change the meaning and is therefore best avoided.

4) **Writing to be ‘tightened’ throughout:** General comment on style, not clear what specifically could be changed. As reviewers 2 and 4 comment favourably on the writing style we have not made any revisions to the style of writing.

**Referee 2: Margaret R Seashore**

1) **Table 1:** We agree that the table content is confusing and have changed the table to be more concise. That these are actual models, planned or already in place, reported by our informants is made clear in the first sentence of the paragraph on ‘description of regional models’ in the results section.

**Referee 3: Bradford L Therrell**

1) **Page 9, last sentence:** This sentence has been changed to make the meaning clearer.

2) **Abstract background:** The word ‘new’ has been removed

3) **Abstract methods:** Analysis included as part of ‘methods’ in abstract

4) Text in general, use screening in preference of ‘tests’: this has been changed where relevant

5) **National guidance on P4:** The national guidance referred to here is referred to in the background section, at the end of page 2 and the full resource referenced (14). We have added a sentence to the ‘background’ to state that there is no distinction between SCD and CF in this guidance and have clarified on page 4 that the ‘national guidance’ interviewees referred to is that which we reference in the background section.
6) **Resource constraints:** We did not collect data on extent of local resources to support imparting of carrier status information by area. That it has been a factor affecting implementation and choice of methods for informing carrier parents emerged from the interview data and specific details were not requested. All regions would have received the same central financial support for implementing screening.

7) **Local consultation:** Examples of stakeholders have been included to clarify that these consultation were with health care professionals with a strategic, managerial and practitioner remit for delivering screening.

8) **P8, 2nd paragraph:** ‘Respondents felt that’ inserted as suggested. These, and all the other data represented as ‘results’ are the views of our respondents and not author opinion.

9) **General comment:** In the last paragraph of the ‘background’ section we state that the work reported here is part of a wider research study also seeking parents and practitioner views, with a link (reference 15) to further information about the wider study.

**Referee 4: Erika Sims**

1) **Previous experience of screening in region:** Information on when universal newborn screening commenced in each region was collected. However, the duration of experience of screening within a particular region was not mentioned by respondents as a factor influencing choice of method for informing and because we were not attempting to explain reasons for choice of method beyond our informants’ opinions, these data are not reported in the paper. To clarify the variability of duration of screening in England, the first sentence in the second last paragraph of the ‘Background’ section now includes reference to duration of CF and SCD screening prior to the national programmes in parts of the country.

2) **Comparison with experience in Wales, Northern Ireland and Scotland:** We have collected information on practice for informing carriers in Wales, Northern Ireland and Scotland and propose to refer to these experiences in our wider study. While our main study commission (see reference 15) does not allow for comparison or data collection in the other UK countries, we will take their practice into account when considering recommendations for future practice in England in our final report. For this paper we chose to restrict our reporting to the English regions only.

3) **Explanations of abbreviations used in Table 1:** Abbreviations have been removed from the revised table.