Co-designed strategies for delivery of positive newborn bloodspot screening results to parents: the ReSPoND mixed-methods study

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Disclaimer: This report contains transcripts of interviews conducted in the course of the research and contains language that may offend some readers.

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Scientific summary

The ReSPoND mixed-methods study

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Scientific summary

Background

Newborn bloodspot screening in England currently covers nine conditions: sickle cell disease, cystic fibrosis, congenital hypothyroidism, phenylketonuria, medium-chain acyl-CoA dehydrogenase deficiency, maple syrup urine disease, isovaleric acidaemia, glutaric aciduria type 1 and homocystinuria (pyridoxine unresponsive). The last six conditions are collectively referred to as inherited metabolic diseases. Annually, almost 10,000 parents of babies born in England are given a positive newborn bloodspot screening result 2–8 weeks after birth. Despite national guidance from Public Health England, positive newborn bloodspot screening results are inconsistently delivered across geographical regions, and many parents are dissatisfied with how the results are communicated to them owing to inconsistency and a lack of information provision. The expansion of newborn bloodspot screening in England means that there is added pressure to provide cost-effective approaches to communicate positive newborn bloodspot screening results.

Given that most infants will be asymptomatic when the parents receive the positive newborn bloodspot screening result, it is vital that communication is carried out carefully to avoid negative effects on future concordance with treatment and relationships with health-care professionals. Concordance and trust are important to ensure timely uptake of confirmatory diagnostic testing and treatment to maximise outcomes for the child. Poor communication can affect parent–child bonding and ongoing parental and social relationships. For this reason, Family Systems Theory was the theoretical basis for this work.

Aim and objectives

The primary aim of the research was to co-design, implement and evaluate new interventions to improve the delivery of initial positive newborn bloodspot screening results to parents.

This study had the following objectives:

- explore current communication pathways for positive newborn bloodspot screening results from the laboratory to clinicians and then to families
- identify and quantify the costs and benefits of the approaches currently used to deliver positive newborn bloodspot screening results to parents
- select two case-study sites (newborn bloodspot screening laboratories) in which to co-design interventions for communicating positive newborn bloodspot screening results to parents
- develop co-designed interventions in two case-study sites for improving the delivery of positive newborn bloodspot screening results using experience-based co-design by –
    - exploring the experiences of parents receiving and clinicians delivering positive newborn bloodspot screening results
    - producing a composite film of key themes or ‘touch points’ from parents’ perspectives
    - enabling parents and staff to identify joint priorities for improving the delivery of positive newborn bloodspot screening results
    - co-designing interventions for the delivery of positive newborn bloodspot screening results
- implement the new interventions in the selected case-study sites
- undertake a parallel-process evaluation underpinned by Normalisation Process Theory
- quantify the resources that are required to deliver the co-designed interventions in the selected case-study sites and compare these with the costs associated with current strategies

- obtain consensus about the need for and potential design of an evaluation study of the co-designed interventions, including –
    - selection of which co-designed interventions to include in an evaluation
    - selection of relevant outcome measures
    - selection of relevant time horizon and resource use data to collect in a definitive evaluation
    - choice of future study design.

Methods

This was a mixed-methods study underpinned by Family Systems Theory using four phases with defined outputs.

Phase 1

Phase 1 involved a national survey using telephone interviews to explore current approaches to the communication of positive newborn bloodspot screening results and to inform the selection of two study sites for phases 2 and 3. Phase 1 consisted of:

- Sampling. A two-stage sampling approach was employed. Participants were first sampled purposively based on their experience of communicating positive NBS results to parents. This was followed by the second stage, snowball sampling, with the first participants suggesting other relevant clinical colleagues.
- Participants. Participants comprised staff ($n = 22$) in all newborn bloodspot screening laboratories in England ($n = 13$) and representatives of clinical teams ($n = 49$).
- Analysis. Quantitative data collected from the closed-ended questions were analysed using descriptive statistics. Qualitative data from the open-ended questions were analysed using thematic analysis using an inductive approach.
- Outputs. The outputs included description and cost of current communication practices, and selection of two study sites for phases 2 and 3.

Phase 2

In phase 2, an experience-based co-design was used in two selected study sites (newborn bloodspot screening laboratories), which served three NHS trusts, to develop interventions for communicating positive newborn bloodspot screening results to parents. Phase 2 consisted of:

- Sampling. A purposeful sample of parents was used to ensure representation of all screened conditions. For staff, a two-stage sampling approach was employed, as per phase 1.
- Participants. Participants included parents ($n = 21$) across the two study sites who had received a positive newborn bloodspot screening result for their child in the previous 3–36 months. Staff ($n = 17$) across the two study sites who were involved in communicating positive newborn bloodspot screening results in the preceding 6 months were also included.
- Analysis. Themes identified from parental interviews were developed into a composite film. The film was used to capture parents' experiences of receiving their child's positive newborn bloodspot screening result and provided rich information to guide the development of the co-designed interventions. Staff interviews were analysed thematically. An inductive approach to data analysis was used and themes were generated using a latent approach to provide a deeper understanding of the approaches used to communicate positive newborn bloodspot screening results to families. Parents and staff used issues highlighted in the film along with priorities from separate staff and parent meetings to identify joint priorities for improving the delivery of positive newborn bloodspot screening results.
- Output. Outputs comprised interventions for communicating positive newborn bloodspot screening results to parents.

Phase 3

In phase 3, we undertook a parallel-process evaluation underpinned by Normalisation Process Theory and an economic analysis of the interventions in the two selected case-study sites. Phase 3 consisted of:

- Sampling and participants. A purposeful sample of parents who had received a negative newborn bloodspot screening result was included ($n = 14$). A purposeful sample of both parents who had received a positive newborn bloodspot screening result for their child but for whom the interventions had not been used ($n = 12$) and parents who had been given their child's positive newborn bloodspot screening result using the co-designed interventions ($n = 8$) was included. A purposeful sample of midwives ($n = 20$) from the two study sites involved in collecting newborn bloodspot screening data was included. A purposeful sample of newborn bloodspot screening laboratory staff ($n = 5$) and clinicians ($n = 19$) who had used the co-designed interventions to process and deliver the newborn bloodspot screening result to parents in the two study sites was also included.
- Analysis. Interviews with parents of children with a positive result who had and had not experienced the interventions and with parents of children with a negative result were analysed thematically. An inductive approach to data analysis was used and themes were generated using a latent approach to provide a deeper understanding of opinions regarding the proposed interventions. Interviews undertaken with staff were subject to framework analysis.
- Outputs. The outputs comprised a process evaluation and economic analysis, including relevant resource use and associated costs, of the interventions in routine practice.

Phase 4

Phase 4 involved determining the final interventions and design of a future evaluation study. Phase 4 consisted of:

- Sampling. A convenience sample of key stakeholders involved or with an interest in newborn bloodspot screening was recruited.
- Participants. Participants comprised key stakeholders, including representatives from Public Health England involved in the newborn bloodspot screening programme, those involved in supporting families following a positive newborn bloodspot screening result (charities and clinicians) and parents who had received a positive newborn bloodspot screening result.
- Analysis. The principles of the nominal group technique were used to reach consensus on future research priorities.
- Outputs. The outputs comprised a design of a future evaluation study and proposed suitable outcome measures.

Results

Phase 1

Assurance of quality and consistency was a priority for all newborn bloodspot screening laboratories. The findings indicated variation in the approaches to communicating positive newborn bloodspot screening results from laboratories to clinical teams. This was particularly evident for congenital hypothyroidism and was largely influenced by local arrangements, resources and the fact that individual laboratories had detailed standard operating procedures for how they work. Obtaining feedback from clinical teams to the laboratory after the child had been seen could be challenging and time-consuming for those involved. However, this is not the primary role of laboratories and is currently undertaken in this way only because alternative systems are not in place. Pathways for communicating carrier results for cystic fibrosis and sickle cell disease could be ambiguous and inconsistent, which, in turn, could hamper the laboratories efforts to obtain timely feedback regarding whether or not the result had been communicated to the family. Communication pathways for positive newborn bloodspot screening results between laboratories and clinical teams could, therefore, be time-consuming and resource intensive.

Phase 2

Findings indicated variation in the approaches to communicating positive newborn bloodspot screening results to parents, largely influenced by the resources available and a lack of clear guidance. Health-care professionals emphasised the importance of communicating results to families in a way that is sensitive to their needs. However, many challenges hindered communication, including logistical considerations, difficulty contacting the family and other health-care professionals, language barriers, parental reactions, resource considerations, lack of training and insufficient time.

Parents indicated the aspects of communication that they perceived to be both helpful and unhelpful. Helpful approaches included ensuring that the professional who communicated the positive newborn bloodspot screening result was knowledgeable about the specific condition, and was positive, supportive, empathetic, reassuring and credible; tailoring and pacing information to accommodate parents' reaction to receiving the result; providing information to parents immediately after communication of the positive newborn bloodspot screening result to reinforce and clarify what parents had been told and what would happen next; and ensuring that the time between communicating the positive newborn bloodspot screening result and seeing the specialist clinical team was as short as possible. Unhelpful approaches included mothers being told the newborn bloodspot screening result when they were on their own and then having to relay the information to their partner; the result being portrayed as bad news; and being told not to use the internet to search for information about the suspected condition.

Four co-designed interventions were developed: proposed changes to the newborn bloodspot screening card; development of condition-specific, standardised laboratory pro formas; condition-specific communication checklists; and an e-mail or a letter template to provide information to parents following the initial communication of the positive newborn bloodspot screening result. Following discussions with the Newborn Bloodspot Screening Programme (Public Health England), the last three interventions were implemented in practice during phase 3.

Phase 3

The co-designed interventions were fully adopted by one newborn bloodspot screening laboratory (serving two NHS trusts) and partially adopted by the other. Feedback regarding the proposed changes to the newborn bloodspot screening card was generally positive and included recommendations for how the changes might be operationalised in practice. Most parents, and staff who had implemented the laboratory pro formas and communication checklists in practice, recognised the potential advantages in terms of standardisation that these offered. Others, mainly those who had not used the co-designed interventions, felt that communication of positive newborn bloodspot screening results was too nuanced and complex to enable it to be standardised. The provision of information for families following communication of a positive newborn bloodspot screening result was generally viewed positively by all. Implementing the co-designed interventions in practice highlighted numerous organisational and contextual factors that influenced their success.

The cost analysis showed that implementing the interventions would not influence NHS expenditure; implementing the interventions during home visits for the ≈ 8152 sickle cell disease carriers per annum and for the 120 cystic fibrosis carriers per annum would increase the NHS cost by at least £617,298 and £10,801, respectively. Nevertheless, using teleconsultations to inform the parents about the sickle cell disease status of their infant could lead to an ≈ £10,794 saving in the case of carriers and ≈ £19,030 in the case of those affected by sickle cell disease. Although this trend was not observed for cystic fibrosis carriers, deploying the intervention by teleconsultation could lead to a saving of ≈ £10,860 in the case of infants affected by cystic fibrosis. Assuming that teleconsultations would not have a detrimental impact on parents' health and would not increase costs, the interventions could be cost-effective.

Phase 4

Consensus regarding the need for and potential design of an evaluation study of the co-designed interventions was reached and suitable outcome measures for a future evaluation trial were proposed. In addition, proposed changes to the newborn bloodspot screening card will be presented to Public Health England for inclusion in its 5-year plan.

Conclusions

There continues to be wide variation between practices across England in communicating positive newborn bloodspot screening results from the laboratory to clinicians and then to parents; this is influenced by practical, organisational and contextual factors. Although there was evidence of good practice, there was also potential for real and repeated harms when communication was poor or inconsistent. In addition, some existing practices were viewed as time-consuming and resource intensive. These included efforts made by laboratories to gather feedback after the baby had been reviewed by the relevant specialist team following the positive newborn bloodspot screening result. However, as this is not the primary role of the newborn screening laboratories, alternative routes, such as clinicians reporting outcomes directly to a central information point, may be more acceptable and more successful for all involved.

The co-designed interventions showed promise in improving communication of positive newborn bloodspot screening results. Feedback from staff indicated that the proposed changes to the newborn bloodspot screening card would be of value in terms of aiding staff communication, but operationalising these was seen as challenging. Furthermore, standardisation of communication from the laboratory to clinical teams and from clinical teams to parents was viewed as advantageous by many, but implementation raised many challenges; many of these were focused around the organisational culture. Providing information to parents following communication of a positive newborn bloodspot screening result was generally viewed favourably, although the best method(s) and format(s) to use still needs to be teased out.

The over-riding message from parents in this study was that there is no justification for variation in practice and that communication of positive newborn bloodspot screening results requires improvement to avoid potential deleterious effects on them.

Recommendations for future research are:

- further feasibility testing, followed by a national evaluation of the laboratory pro formas, communication checklists and information provision for parents following a positive newborn bloodspot screening result, including proposed outcome measures
- exploration of parental needs during the 90 days following a positive newborn bloodspot screening for sickle cell disease
- identification, assessment and evaluation of models of care for communicating positive newborn bloodspot screening results for congenital hypothyroidism.

Trial registration

This trial is registered as ISRCTN15330120.

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