

Provision of information about newborn screening antenatally: a sequential exploratory mixed-methods project

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Scientific summary

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Background

Newborn (neonatal) bloodspot screening (NBS) involves collecting a blood sample from a newborn baby aged between 5 and 8 days. In England, NBS is introduced to parents as a recommended routine screen for nine conditions, but assumes an informed consent model. Ensuring that parents are adequately informed is the role of midwives, supported by a screening booklet. NBS communication and consent processes are complex and there is no evidence-based standardised model. There is, however, increasing appreciation of the centrality of communication in ensuring that the maximum benefits of NBS are realised, while reducing potential harms. Specifically, there is a recognised need to inform parents prior to screening, preferably antenatally. However, NBS information represents one of the largest volumes of screening information that people might assimilate during their lifetime. The need for effective communication has increased as a result of an expansion in the number of conditions screened for in NBS panels, including those for which there are less clear treatment benefits. The additional cost of expanded NBS, potentially requiring extra input from midwives and removing them from other duties, needs to be worth the potential benefits for parents and babies. These benefits may include both health and non-health benefits, such as the value of additional information per se that comes from screening results and the avoidance of distress if parents are prepared for screening. At present there is insufficient evidence to effectively design a comparison study of alternative communication models suitable for NBS.

Aims and objectives

This study aimed to determine service providers' and users' views about the feasibility, cost, efficiency, impact on understanding and the consent process of current practice and preferences for alternative methods of conveying information antenatally in a Newborn Bloodspot Screening Programme (NBSP).

The study had nine objectives:

1. collate the characteristics of potential alternative communication and consent models for NBS
2. explore how providers and users envisage that information given antenatally can best meet the challenge of effectively and efficiently providing parents with sufficient understanding of an extended NBSP
3. examine parents' understanding and experiences of NBS communication to draw inferences regarding best practice within extended NBS
4. establish the resource use and costs associated with the current practice of providing NBS information
5. examine the preferences of midwives and current and future parents with regard to information provision in a NBSP
6. establish the key parameters affecting the cost-effectiveness of new modes of information provision compared with the current practice of information provision in a NBSP
7. outline the key uncertainties in the current evidence base and quantify the value of future research for evaluating the effectiveness and cost-effectiveness of providing NBS information
8. explore providers' and users' views on the study suggestions, focusing on acceptability, broader impact, effectiveness, efficiency and parent understanding
9. establish how generalisable the study findings are across conditions screened for in the NBSP.

Methods

A realist review and in-depth qualitative interviews were used to generate alternative communication and consent models informed by international evidence and health professionals' and parents' experiences. A telephone survey and an observation study were used to identify the resources used in, and costs related to, current practice. A stated preference survey was used to quantify preferences for information provision within a NBSP. An early model-based economic analysis was used to identify the key drivers of the relative cost-effectiveness of potential modes of information provision and the value of future research. Focus groups were then used to explore the implications of the project findings from the viewpoint of parents, service providers and key stakeholders.

Results

Study 1

Key concepts within an analytical framework of consent, which followed the dominant autonomous authorisation model, were relevant information, understanding, retention, intention, intellectual capacity and not being coerced. Empirical studies ($n = 47$) within the NBS communication and consent literature were synthesised into this framework. Literature prominently focused on *relevant information* and that parents should *not be coerced*, with a paucity of evidence about *understanding* and *retention* and an absence of data to inform *intellectual capacity* and *intention*. Questions were raised about whether or not autonomous authorisation is a suitable model for NBS. There was no evidence supporting a uniform effective communication model but key elements were that the timing of communication is pivotal, with post-birth communication being particularly ineffective; health professional communication is preferred and more effective, but this could in part be because of serious current limitations with alternative modes of information provision; and consensus on key topics is achievable, but parent selection of both the level of information and the mode of information is desirable and likely to be more effective. A pre-interview tool was produced from this review to help understand the views of interviewees about alternative communication and consent models.

Study 2

Interviews with 45 parents and 37 health professionals representing midwives and NBS co-ordinators found that parents wanted NBS information 'drip fed' throughout pregnancy. All interviewees thought that 36 weeks' gestation was a key time when information on the child became relevant. Post-birth reminders were needed, but all agreed that new information should not be given then. Both groups favoured tailored 'personalised' information to suit individuals' learning and information needs, with parents choosing the format suitable for them ('parent led'). Applications (apps) and websites were favoured by parents if they contained general pregnancy information in the early postnatal period. Parents supported the use of e-mail and text notifications and clinic television to remind them to engage with NBS information, highlighting an opportunity for the NBSP to use current technological routes that parents use for gathering other information. Both samples thought that antenatal classes and leaflets were useful for subgroups of parents. All parents saw a brief discussion with a midwife as being essential for introducing screening and signposting the range of information resources available.

Most parents whose child had been screened felt that screening had not been presented to them as a choice. However, most were happy for midwives to assume their consent if they were adequately informed. Midwives also thought that it was important that parents were fully informed; however, they acknowledged that it was difficult to obtain true informed consent in practice.

Most parents had a very low awareness of NBS even after screening. Most parents, with the exception of participants in the medical profession, lacked knowledge about the conditions being screened for, prior to and after screening had taken place. Furthermore, many parents did not fully understand why their child had been screened. Often, their understanding of information had not been checked prior to NBS.

Parents repeatedly stated that they had not realised how serious NBS outcomes could be and compared this with the higher levels of knowledge that they had for other screening that occurs antenatally. Parents felt that their lack of awareness of NBS and engagement with NBS information was because NBS was presented to them as a routine part of care, because of the inappropriate timing of information provision and because NBS information was provided along with large amounts of information about other health initiatives or other promotional materials. Parents and midwives had concerns that mothers whose first language was not English were not receiving information in their primary language. These data suggest the need to optimise information provision timing to ensure that communication is effective, make parents aware of the relevance of NBS information, ensure that information is accessible in multiple formats and levels, and ensure that parents are aware that screening is optional and not part of routine care.

Study 3

This study identified the key items of resource use and potential cost drivers in current practice using the perspective of NHS England. The time frame for the analysis started from the time of initial information provision to the time at which informed consent was obtained. Using the data collated from the telephone interviews (22 midwives) and observation study (nine observations), the estimated expected cost per parent of NBS information provision was £17.65. This can be extrapolated to a total cost of £11,675,404 to NHS England per annum, using the estimated number of babies born in 2014. There is likely to be significant uncertainty in this estimate of the cost of information provision in NBS using the current approach of information provision by midwives with the support of leaflets.

Study 4

A hybrid stated preference study using a linked conjoint analysis–discrete choice experiment quantified the preferences of samples of the general public representing the views of current and potential future parents and midwives for information provision in NBS in England. Parents' and midwives' preferences relating to NBS information provision differed from current practice. Realignment of information provision to match these preferences could help improve parents' ability to make screening decisions and help midwives focus what information is provided. These data suggested that one potential NBS communication model may be for midwives to provide information verbally after the 20-week scan.

Study 5

A systematic review of published economic evaluations of NBS identified that information provision costs and the effect of poor information provision on parents are rarely included in evaluations. An early model-based economic analysis examined what resource use, costs and outcomes may be affected when using modes of information provision in the context of national NBS. A decision tree compared two new information provision modes with two types of current practice, reflecting how information is currently provided and how information should be provided if current information provision guidelines are followed. The economic analysis assumed the perspective of a health-care system and reflected information provision to the mother from when the baby is conceived up until the final NBS result is received. Because of a paucity of data to populate the model, the absolute expected costs and benefits from the economic analysis should be interpreted with caution. This analysis indicated the relevant items of resource use, costs and outcomes that are likely to influence the relative cost-effectiveness of different information provision modes and suggested that further research would be of value.

Study 6

The stakeholder consultation study involved 12 health professionals and five parents, who participated in focus groups and telephone interviews. Parent and health professional views were largely in agreement. Both groups were highly supportive of an 'information tool kit' to personalise information provision and of focusing health professional communication in the last trimester. A tool to enable midwives to highlight changes in screening since a previous pregnancy was supported. This approach was seen as a more effective way of ensuring parent understanding and use of midwives' time. Parents withdrew support for mandatory screening once they were aware of bloodspot storage. They also highlighted how receiving key NBS information after screening has occurred can erode trust and the public view of NBS.

A sample consisting of midwives, senior members of the Royal College of Midwives, representatives of the cystic fibrosis screening programme, a NBS laboratory director and a parent of a child identified as a sickle cell carrier was used to explore the potential generalisability of the study findings. The data suggested that the communication and consent challenges were not condition specific. The findings also highlighted the potential impact of parents not being aware that their child could be identified as a carrier and yet the low priority health professionals gave conveying this message.

Conclusion

The novel findings from this study have resulted in further understanding of NBS communication and consent. The cost of providing information across England using the current approach was estimated to be £11,675,404 per annum, representing a substantial cost relative to the current estimation of NBS costs. Service providers and users questioned whether or not the goals of communication, to prepare parents and to gain informed consent, are currently being achieved. These goals do appear attainable, however, with the findings suggesting a preference for a personalised approach to NBS information provision, with information and consent prioritised in the third trimester.

Implications for health care

- Given the variation in parents' information needs and opinions on how best to receive information, there is a need to develop a personalised approach to information provision including formats that include core topics but enable parents to seek different levels of detail.
- Midwives should focus on checking parents' understanding rather than being the sole providers of information.
- Increased NBS awareness, specifically that parents are expected to make a decision, was desired and could be achieved through a number of mechanisms that parents already use to gain information.
- The timing of information provision was identified as one of the most influential factors determining its effectiveness. Information provision after birth is particularly ineffective. The third trimester was a key time for information assimilation.
- Although midwives are not currently allowed to distribute any advertising materials, it was clear that parents felt that the health information given in pregnancy often has to compete with marketing information for their attention. Parents wanted guidance on what information to attend to and wanted important health information to be clearly marked.
- Despite translated materials being available, midwives and parents raised concerns about the quality of information provision for women who do not speak English as their first language.
- Information was valued by some more than choice, with parents generally supporting mandatory or assumed consent. However, health professionals preferred informed consent, as did parents who became aware of the storage of samples.
- Currently, messages about false-positive results were undervalued by health professionals and seldom given, while the potential for carrier results was not seen to be discussed in the observation study. Yet preparedness for these events can limit psychological distress and the need for specialist service use. Designing NBS information around recipients' information needs across the screening pathway, rather than focusing on consent, may enhance effectiveness.

Recommendations for research

- The development and robust evaluation of an information tool kit to provide a personalised approach to information provision in NBS through a trial of the personalisation mechanism and 'information tool kit' offered to parents in the third trimester of pregnancy compared with current actual practice.
- The development of a mechanism to target the components of the information tool kit to subgroups of parents such that the mode of communication matches parents' attitudes (active or passive) to information provision.

- Use this trial of the information tool kit to examine the effect of changing the role of midwives from providing a list of information to becoming a signposter and checker of parental understanding.
- Use the current practice arm of the trial to understand why health professionals currently provide information at the time and in the way that they do.
- Identify the appropriate primary outcome measure to capture the effectiveness of the information tool kit for parents taking part in a NBSP.
- Quantify the impact of different approaches to information provision on anxiety levels in parents.
- Quantify the impact of different approaches to information provision on the use of health-care resources and, specifically, identify the level of uptake of visits to health-care professionals and the NHS when a positive result has been received and a confirmatory test result is awaited.
- Develop an understanding of how to trigger understanding rather than focusing on information provision.

Trial registration

This trial is registered as ISRCTN70227207.

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