PUBLIC HEALTH 178 (2020) 151–158

Available online at www.sciencedirect.com

Public Health

journal homepage: www.elsevier.com/puhe

Original Research

Consent for newborn screening: screening professionals' and parents’ views

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ARTICLE INFO

Article history:
Received 18 June 2019
Received in revised form
23 July 2019
Accepted 15 August 2019

Keywords:
Newborn screening
Consent
Midwives
Parents
Qualitative methods
Decision

ABSTRACT

Objectives: Expansion of newborn bloodspot screening (NBS) within England, which practices an informed consent model, justified examining acceptability and effectiveness of alternative consent models.

Study design: Qualitative focus groups.

Methods: Forty-five parents and 37 screening professionals (SPs) participated. Data were analysed using thematic analysis.

Results: Parents and SPs initially appeared to have differing views about appropriate consent models. Most parents accepted assumed consent, if adequately informed; however, once aware of bloodspot storage, informed consent was wanted. SPs valued informed consent, but acknowledged it was difficult to obtain. Both samples wanted parents to be informed but were unclear how this could be achieved. Most parents felt NBS was not presented as optional.

Conclusion: The simultaneous exploration of parents and SPs views, in real time is original. This rigour avoided the reliance on retrospective accounts which make it difficult to establish how decisions were made at the time. It is also unique in providing pre-interview consent models to drive the depth of data. It was rigorous in member checking. Findings suggested a preference for full disclosure of all information with some parents valuing this more than choice. Both samples queried whether current consent was sufficiently informed and voluntary. Results suggest differing tolerances of consent type if screening is solely for diagnostic purposes vs bloodspot storage. Results highlight the need for caution when examining consent model preferences without also checking knowledge, as opinions may be based on incomplete knowledge. Future research is needed to examine efficacy of proposed changes.

Funding: National Institute for Health Research Health Technology Assessment HTAProgramme (11/62/02).

Trial registration: ISRCTN70227207.

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https://doi.org/10.1016/j.puhe.2019.08.009

Introduction

Newborn screening involves taking samples of blood from a baby's heel (the bloodspot test) to screen for serious conditions in babies within the first days of life. Early identification enables timely treatment, leading to improved prognosis and quality of life. However, consent for newborn bloodspot screening (NBS) is complex. Consent models differ internationally ranging from mandatory screening—the state decides to screen; ‘opt out models’—the state recommends but parents can decline to voluntary informed consent—the state recommends, but parents indicate willingness for screening to be performed. Where consent is needed, parents are deciding on behalf of their newborn—known as proxy consent. The information which needs to be understood is novel and vast; there are numerous rare diseases (9 conditions in England) which are unknown to many,[1] there are a range of possible screening results: diagnosis, inconclusive diagnosis, carrier, suspected carrier, false positive and normal. The results can have implications for family members' genetic risk and paternity, and bloodspots may be stored for anonymised research. Finally, consent is taken 5 days after birth when parents are tired and processing volumes of information regarding their child. Concerns have been raised about the validity of consent for NBS in practice as uptake is high in most countries,[2–7] yet parental awareness and knowledge levels are low.[8–12, 30, 34, 35, 36, 37, 38, 39, 40]

NBS in England is promoted as only performed after receiving valid informed consent from parents[13] but is offered to parents routinely. Midwives inform parents and distribute a screening booklet. However, this high standard of consent can be difficult to achieve as outlined above. Two Health technology assessment (HTA) studies have reported concerns regarding whether parents in England were adequately informed before screening.[15,16] If decisions about screening are not sufficiently informed, consent validity is questionable. This study sought to examine parent and screening professional (SP) preferences for different NBS consent models.

Methods

Participants

As previous work[17] evidenced variation in Newborn screening programme (NBSP) communication practice which will affect views of consent, all Senior Quality Assurance Managers in England were sampled to capture maximum practice variation ($n = 6$). Through these a regional quality assurance manager ($n = 1$), hospital screening coordinators ($n = 13$), community midwives ($n = 14$) and hospital midwives ($n = 3$) were sampled proportional to NBS involvement and purposively to capture variance in experience and regional coverage. The SP sample ($N = 37$) covered 16 trusts in rural and urban areas of England. All regions of England were represented.

Parents were sampled across the screening pathway: parents-to-be ($n = 14$), parents ‘screened waiting for result’

Research in context

Evidence before this study

Countries vary in the consent model used. In countries with informed consent models, concerns exist about the validity of consent. Although all countries value informing parents about screening, there is a lack of an effective model. The increase in the number of conditions included in newborn screening raised questions about what consent model is most appropriate.

Added value

This is one of the first studies to explore the views of parents and screening professionals simultaneously and in real time. Previous research has either separated these samples, therefore, because newborn bloodspot screening (NBS) is constantly evolving, parents and health professionals may be providing views on different forms of NBS, or it has relied on retrospective parental accounts or subsamples of parents affected or professional groups. This 360 design has enabled contrasts to be made between participants and the unpicking of what is driving opinions. Furthermore, its novel design of a pre-interview contemplation exercise enabled more in-depth data to be gathered, enabling the team to understand views which are likely to drive responses to NBS. The design highlighted the risk of creating policy on research without a clear idea of participants knowledge and the damage to public trust that can be done when parents become aware of key screening information after screening has been performed. The above not only adds to the evidence around optimum prescreening information provision but also can help explain why parents may have such negative responses to positive screening results.

Implications of all available evidence

Both parents and screening professionals value parents being informed so highly that it drove their preferences for different consent models. Indeed information was more valued than choice for some.

Parents hold newborn screening in high regard, but this may be based on a belief of screening confirming a state of health and are often are unaware of their choice or of other potential outcomes or bloodspot storage. This high regard and the use of leaflets may be driving low parental engagement with screening information, whereby screening is performed because of parental trust in the NHS.

Designing materials which prioritise information about choice and bloodspot storage may increase parental engagement with information and help to achieve the goal of parents feeling sufficiently informed.