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Developing a Stated Choice Experiment to Understand Preferences for Information Provision in an Expanded Newborn Bloodspot Screening Programme

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Key messages

- A hybrid stated choice experiment was designed to elicit parent and midwife preferences for newborn bloodspot screening (NBS) information provision.
- The survey comprised a conjoint analysis linked with a discrete choice experiment
- The outcome of information provision 'being able to make an informed decision' was used as the linking attribute
- The next phase is to pilot the survey design in a larger sample of parents (current and future) and practising midwives

Introduction

Newborn bloodspot screening (NBS) is a public health programme which aims to detect severe inherited illnesses in newborn children. By identifying children with such conditions at an early age, treatment can be given which can help to prevent or reduce the health implications to the child. In the UK, participation in the NBS should only occur after parent(s) have provided informed consent. However, evidence suggests that many parents have a poor understanding of NBS, with many not knowing "specifically what the testing had been for"¹. Such lack of information can cause future problems such as parental anxiety and unnecessary health care resource use². Given that the UK NBS is due to expand from the current five conditions to screening for nine conditions, these problems may only grow in magnitude.

Understanding parents and midwives' preferences for aspects of newborn bloodspot screening would allow this information to be tailored so as to allow it to be better

taken on board. In theory, appropriately tailored information provision would reduce parental anxiety experienced on receipt of positive results or when a repeat test is needed. The experience and opinions of midwives must also be incorporated when designing such an intervention. This is particularly true as any additional time spent providing information will impact on their other duties or result in them working for longer hours. By balancing the preferences of both of these groups, a resource-effective programme of information provision can be constructed which prepares parents for screening and its possible consequences.

Aim

To design a stated choice experiment (sCE) to elicit parent and midwife preferences for information provision in an expanded NBS programme in the UK.

Method

Mixed methods were used to define the (i) choice question (ii) type of sCE (iii) attributes and levels.

Four methods were used:

- A systematic review of published (2001-2014) discrete choice experiments (DCEs) eliciting preferences for screening, diagnostic or genetic tests or maternity-related services. The attributes and levels identified from published DCEs were tabulated and potentially relevant examples extracted,
- A rapid review of NHS-NBSP policy and practice documents and training materials. Key characteristics that are reported to be a requirement of information provision in NBS were listed.
- Consultation with NHS-NBSP expert. The list of potentially relevant information attributes was discussed with experts.
- Semi-structured interviews with parents and regional quality assurance coordinators. Qualitative data elicited from interviews were analysed using thematic analysis.

Results

Systematic Review

A total of 58 relevant DCEs were identified and unique attributes were tabulated for each type of intervention. Duplicate attributes were assimilated and the remaining unique attributes were grouped into 20 common themes. Themes which were not relevant to the policy context were then removed leaving 13 attributes, with 11 being represented in the conjoint analysis (part A of the survey) and 3 in the DCE (part B of the survey).

Rapid Review

Three key newborn screening documents were reviewed to aid the development of the sCE. These were:

- the current "Blood Spot Screening for You and Your Baby" leaflet,
- a set of postnatal notes which are given to parents and contain some information and
- a handbook designed for health professionals who take the bloodspot sample³.

These documents were used to validate the attributes to be included in part A (reflecting types of information required) with the current screening leaflet representing current practice. The health professional handbook also helped to reinforce current practice.

Qualitative interviews and Consultation with Experts

Semi-structured interviews have also been used to inform the design of the sCE. Participants in these interviews included 20 parents and 7 regional quality assurance co-ordinators in newborn bloodspot screening.

Two senior academic midwives, 4 screening co-ordinators, 2 laboratory scientists, and a patient-support charity also offered their opinions on the design of the experiment, although not within the same formal interviews.

Emerging qualitative themes indicated: (i) the complexity of the choice question; (ii) the relevant study population (new parents; members of the public of child-bearing age; midwives); and (iii) three key aspects needed in the sCE design:

- The extent and type of information attributes to facilitate ability to make an informed decision
- Process attributes related to approaches to information provision relative to the ability to make an informed decision (outcome attribute)
- Capturing the impact of expansion in number of conditions tested by using two survey versions.

These qualitative data suggested it was also necessary to use a measure to capture respondents' views about shared decision making in the context of NBS. In this survey an adaptation of a published measure will be used⁴.

Together the results suggested that a hybrid-conjoint analysis (part A – see Table 1) with DCE (part B – see Table 2) was the appropriate type of sCE. The two types of stated preference survey would be linked using the outcome of information provision 'being able to make an informed decision'.

Attributes ^a
Do I have a choice about whether to take part in the NBS service?
What are the names of the conditions included in the NBS service?
What can I do to prepare my baby for when the sample for the NBS service is taken?
How will the sample for the NBS service be taken?
Will my child suffer any pain or side effects when the sample for the NBS service is taken?
How long I have to wait until I receive the results?
How I will receive the results?
Does the NBS service identify every child who has one of the conditions?
Is it possible that the NBS service could suggest that my child has a condition when in reality they do not?
What effect would having one of the conditions have on my child's health?
How common or rare are the conditions?
If my child has one of the conditions, what can be done to treat them?

Table 1: Attributes included in the conjoint analysis

^a Each attribute is assigned two levels (yes – information is available to answer this question; no – information is not available to answer this question)

Attribute label	Levels	
When in the pregnancy information is provided	Early pregnancy	The information will be given early in the pregnancy at the first booking appointment (< 28 weeks)
	Late pregnancy	The information will be given late in the pregnancy after the 20 week scan but before the baby is born
	When the baby arrives	The information will be given when the baby is born on discharge from the hospital or a midwife led unit
	Post-birth (day 3)	The information will be given after the baby is born and 24 hours before the sample will be taken
	When the sample is taken	The information will be given immediately before the sample is taken
How the information is provided	Discussion	The midwife will describe and discuss the NBS service with the parent(s)
	Leaflet	The midwife will give the parent(s) a leaflet containing the information.
	The Internet	The midwife will tell the parent(s) where they can find the information about the NBS service on the internet
	Mobile/Tablet App	The midwife will tell the parent(s) how they can download an app which contains information personalised to them
	Group session	The midwife will tell the parent(s) the information as part of a group session with other parent(s)
Parent (s) involved in shared decision making with the midwife	yes	One or both parents are involved in making the decision to take part in the service with the midwife
	No	The midwife does not involve either parent in making the decision to take part in the service
How does the information provided affect parent (s) ability to make a decision	The rating is measured on a scale	1 = It is very hard to decide 2 = It is hard to decide 3 = It would not affect my decision 4 = It is easy to decide 5 = It is very easy to decide
The cost to the parents	£5	These values represent one off costs that may cover, for example the time and travel costs for obtaining the information as part of the NBS service
	£25	
	£50	
	£75	
	£100	

Table 2: Attributes and levels in the discrete choice experiment

Conclusion

This study developed a hybrid sCE to provide a practical solution to understanding preferences in complex situations. This was achieved by separating preferences for processes and outcomes into a conjoint analysis and sCE and including the individual's preference rating for the former as an attribute in the latter.

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