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Eliciting Preferences for Information Provision in Newborn Bloodspot Screening Programs

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ABSTRACT

Background: The national newborn bloodspot screening programs (NBSPs) are continually expanding to screen for more conditions. **Objectives:** To quantify parents' preferences for information and the way in which this is provided in example NBSPs. **Methods:** A hybrid choice experiment, combining a conjoint analysis and a discrete choice experiment, was designed. A sample of current and future parents between the ages of 18 and 45 years was identified via an Internet panel. Respondents completed one of two survey versions (9 conditions and 20 conditions) comprising a validated measure of attitudes toward involvement in decision making, 6 CA questions (11 information attributes), 10 DCE questions (4 attributes: 3 process and the ability to make an informed decision), and demographic questions. **Results:** Of the 702 respondents who completed the survey, 58% were women, 48% were between 25 and 34 years old, and 48% were current parents. All types of information were identified to statistically significantly improve parents' ability to make a decision.

Participants preferred taking an "active" role in decision making. Respondents to the 9-condition survey preferred information before 20 weeks (willingness to pay [WTP] £11.88; CI £5.56 to £19.53) and the 20-condition group after 20 weeks (WTP £15.91; CI £10.64 to £21.63). All respondents disliked receiving information 3 days after birth, with the 20-condition group also being averse to receiving it on day 5 (WTP –£11.20; CI –£18.40 to 5.72). Respondents in both groups preferred to receive their information in an individual discussion. **Conclusions:** This study suggests that parents' preferences for receiving NBS information differ from how this information is given in current UK practice.

Keywords: conjoint analysis, discrete choice experiment, information provision, newborn screening, outcomes, preferences, process.

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Introduction

Newborn bloodspot screening programs (NBSPs) have become established as national programs in countries across the world and aim to identify the presence of a range of inherited conditions in newborns [1]. The number and type of conditions included in NBSPs differ by country, and in some instances, within country, such as in the United States [2]. In the United Kingdom, the NBSP has recently expanded to include nine conditions, having previously screened for five conditions: phenylketonuria, congenital hypothyroidism, cystic fibrosis, sickle cell disease, and medium chain acyl coenzyme-A dehydrogenase deficiency. The four new conditions added were maple syrup urine disease, isovaleric acidemia, glutaric acidemia type 1, and homocystinuria [3].

Within an NBSP, the process of screening involves a health care professional, usually a midwife, taking spots of blood from the heel of the newborn within the first week of life. Screening is

conducted early in the child's development with the aim of starting treatment for identified conditions as early as feasible to reduce, or sometimes avoid, damage to the child's health.

In the United Kingdom, participation in the NBSP is voluntary. Midwives, who typically conduct screening, are required to obtain verbal informed consent from parents in order for a child to be screened. In some jurisdictions, such as in specific states in the United States, participation in the NBSP is mandatory, but parents are still likely to gain value from information about the process of screening and the role of the program. There is substantial evidence to support that after the screening, parents often do not recall what the screening was for, its implications, or that it even took place [4–6]. Furthermore, evidence suggests that a lack of understanding about screening can lead to heightened parental anxiety on the receipt of positive, carrier, or equivocal results [6–8]. It is also possible that a lack of a clear understanding about the role of the program will impair parents' ability to make informed decisions, which may lead to parents using a

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disproportionate amount of health care resources given their concern for their child [9,10].

As NBSPs continue to expand to include more conditions [3], it is likely that it will become more difficult to ensure that parents are informed sufficiently about screening for each condition. Alongside the challenge of what information to provide, another process-related aspect is how to provide the information, particularly in a resource-constrained environment such as in an NBSP in which midwives have only limited time to communicate with parents and take the required sample. In this context, the relevant outcome may also extend beyond simply capturing gain in health status to being able to quantify if, and how, the information provision has affected parents' capability to make an informed decision [11]. Current evaluations of NBSPs and screening technologies rarely account for the costs and benefits associated with the need to inform parents about screening and the consequences of failing to provide adequate information [2]. When these factors were accounted for, providing information was found to pose a significant monetary cost for the health care provider, and poor information provision was identified as a factor that could exacerbate the length and degree of anxiety caused by the screening process. A prolonged and raised anxiety level may be a factor that impairs parents' decision making, leading to higher health care resource use.

Two existing studies have been designed to elicit preferences in the context of NBSP. Miller et al. [12] found that members of the public placed the highest value on a screening program that provided the most clinical health benefit, whereas reproductive risk information and early diagnosis were also valuable characteristics. Hendrix et al. [13] investigated the preferences of black and low-income parents, a section of the public under-represented in research, about the use of dried bloodspots in a research context. This study found that the most important factor driving preferences was that parents felt they should be asked to give consent for the storing of bloodspots. Other important characteristics were whether the child could be linked to the bloodspot and the affiliation of the researchers using the bloodspot sample. To date, no study has identified which information is preferred by parents and how this is provided in the context of an NBSP.

This study aimed to elicit the preferences of current, and future, parents. The study had three specific objectives to identify: 1) what types of information are preferred when parents make the decision about whether their child should participate in an NBSP; 2) how information should be provided; and 3) the balance between the ways in which information is provided, with parents' ability to make a decision about participation in screening.

Methods

The research question being addressed was "What types of, and how should, information be provided, when compared with ability to make an informed decision to participate in the NBSP?" This research question required the elicitation of preferences for a number of information, process, and outcome attributes. To accommodate a substantial number of attributes it was necessary to use a hierarchical information integration (HII) experiment [14–19]. This type of stated preference survey uses subexperiments to value attributes that are grouped in a logical way. These groups are then traded off against one another to find their relative values. The value of items within each subexperiment can then be inferred. This has the advantage of allowing a large number of heterogeneous attributes to be valued. An assumption, however, must be made that choices in each subexperiment are independent.

A hybrid survey design comprising a rating-based conjoint analysis (CA) and a separate, but linked, binary discrete choice experiment (DCE) with an opt-out was used to quantify the preferences of a representative sample of the public for the process and outcomes of information provision in NBSPs. Using a CA, with a rating scale for respondents to indicate their preference, and a DCE, in which respondents tick the preferred option, required the use of a "bridging" attribute that linked the two components of the survey. The rating scale used in the CA asked respondents to assess how the types of information affected the "ability to make an informed decision" in the context of an NBSP. This was then linked with the DCE because the ability to make an informed decision was an attribute included in the DCE. The study was designed in accordance with published guidelines for the design of studies using CAs [20] and DCEs [21]. Ethics approval was obtained from the University of Manchester Research Ethics Committee.

Identifying Attributes and Levels

Mixed methods were used to identify and frame the attributes and levels for both the CA and the DCE. All attributes in screening-related DCEs ($n = 58$), identified from a broader systematic review of published DCEs [22,23], were tabulated to generate a list of attributes ($n = 13$). Eleven of these attributes represented the type of information and were included in the CA. Two of these attributes were related to the process of information provision and were included in the DCE. Three screening policy documents were used to validate the attributes to be included in the CA (reflecting types of information required) with the current screening leaflet representing current practice [24–26]. Semistructured interviews with parents ($n = 20$), midwives ($n = 29$), and NBSP regional quality assurance managers ($n = 7$) did not identify any new potential attributes but suggested that it was necessary to try to elicit how preferences for information may change given the number of conditions included in the NBSP. The interviews also suggested there was a clear trade-off to be made between the number and type of information preferred, impact on ability to make an informed decision, and time available to the midwife to provide information.

The findings from each of these studies were triangulated [27] to identify areas of dissonance, agreement, or silence and create a "short list" of potential attributes and levels. Consultation with National Health Service NBSP experts ($n = 3$) confirmed the relevance of these attributes and levels. A set of attributes and levels for the CA and DCE was then piloted using think-aloud methods in a sample of five members of the public and university researchers and analyzed using content analysis before the final attributes and levels were selected and framed. Using face-to-face survey completion of a mock-up survey it was identified that the type of condition being screened should not be included as an attribute because this would correlate directly with the attribute in the CA that asked respondents whether they wanted to know what conditions were included. Subsequent piloting confirmed that a feasible approach to look at the impact of the number of conditions in the NBSP was to create two survey versions for an NBSP that screened the baby 1) for 9 conditions (as is the current practice in the United Kingdom) and 2) for 20 conditions, representing a theoretical expanded NBSP. Tables 1 and 2 present the final lists of attributes and levels for the CA and DCE, respectively.

Experimental Design

The CA was designed using an orthogonal main effects design to allow the unbiased estimation of main effects among types of information [28]. This design created a total of 24 questions that

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