Informed Choice for Newborn Blood Spot Screening in the United Kingdom: A Survey of Parental Perceptions

Objective: In the United Kingdom, newborn blood spot screening proceeds on the basis of consent based on an informed choice. However, little is known about parent experiences of this process. This study was intended to explore parents’ understanding of newborn screening and their experience of the consent process using a structured survey.

Methods: A mail survey exploring key components of consent was sent to parents whose children had been screened in the year before the study. A descriptive analysis of responses together with logistic regression was performed to identify variables that predicted parental likelihood to agree that they had made an informed choice to screening.

Results: Most parents indicated they understood why the heel prick was done, but only a third understood how conditions would be dealt with if found. Almost a third of parents reported that they did not feel they had a choice to decline screening, yet 70% felt they had made an informed choice. Logistic regression indicated previous children, understanding why the heel prick was done, having time to make a decision, and feeling they had a choice were significant predictors of feeling an informed choice had been made.

Conclusions: The findings support previous studies that screening may be seen as a fait accompli. Analyses indicated that allowing time to make a decision (eg, by providing information during pregnancy, emphasizing the decision-making aspect, and clearly articulating the reasons behind screening) may help parents feel that they have made an informed choice. Pediatrics 2012;130:e1527–e1533
Newborn blood spot screening is a long-standing program through which newborn babies are screened for a variety of conditions shortly after birth. Established internationally, programs now exist in Europe, North America, South America, and the Asia Pacific regions.1–7 There is, however, variation with regard to the delivery of screening programs, particularly in relation to consent. In the United States, screening is generally mandated, although most state programs allow for the refusal of screening on religious or personal grounds.8–11 Other countries, such as Canada, have provincial programs that generally operate on an opt-out basis.12,13 In contrast, newborn screening in the United Kingdom proceeds only after consent based on an informed choice (“informed consent”) has been provided.14,15

As part of the UK national screening program, every resident newborn baby, and those under age 1 year who enter the United Kingdom, should be offered blood spot screening.16 This achieves almost universal uptake.17–20 The blood spot sample is taken between days 5 and 8 of age (usually on day 5) by the midwife, commonly in the infant’s home. Before screening, health professionals (again, primarily the midwife) are expected both to provide written information in the form of the “Screening Tests for You and Your Baby” booklet* and discuss newborn blood spot screening.21 The informed consent process requires health professionals to “Explain fully to parents and then record in the maternity record that newborn blood spot screening has been discussed and recommended, booklet given and consent sought.”22

Consent may be considered informed only if a competent individual voluntarily makes a decision on the basis of disclosed information that is understood.22 Some authors have questioned whether these criteria are being met in the context of newborn screening and cite the high uptake rates as being indicative of testing that may not be proceeding on the basis of informed consent.23 Several studies support this assertion, with parents indicating that they feel that screening was a fait accompli.24–26 Indeed, there is a body of research that indicates parents are often unable to recall specific items of information about newborn screening.27–30

However, much of this work is qualitative, and there is a lack of data as to how common these experiences are. Moreover, a failure to recall information may not be indicative of understanding.31,32 Indeed, the converse may be true: the ability to recall risk information does not necessarily indicate that the individual understands the implications or consequences of that risk.33,34 Recent qualitative work has also suggested that although parents may not recall specific information, they may exhibit an understanding of the motivations for screening and the procedures,32 which may be sufficient evidence of understanding required of an informed consent.35

Consequently, we sought to collect quantitative information regarding parental experiences of the different aspects of informed consent in the context of newborn blood spot screening in the United Kingdom.

METHODS

The population selected was that served by the clinical biochemistry department at the Royal Liverpool Children’s Hospital in their role as part of the UK Newborn Screening Laboratories Network, an association of National Health Service laboratories that provide screening services to newborn babies. The regional newborn screening laboratory hold records of all infants born in the Merseyside and Cheshire regions. Parents were eligible for inclusion if the blood spot from their child had been analyzed during 2008 (n = 28,348). Of these, a random sample of 500 parents was selected. Parents of children who had subsequently died were excluded because it was deemed that the discussion of newborn screening might have caused distress.

Identification and Recruitment

Because records only included maternal contact details, mothers were approached in writing by a clinician (KWS) with appropriate clinical access and received a copy of the questionnaire, information sheet, cover letter, and prepaid envelope. The information sheet also gave mothers a web address for an online version of the survey. Participants were given a small financial incentive to complete the questionnaire. If after 2 weeks parents had not responded, a reminder letter, together with an additional copy of the questionnaire, was sent. Ethical review deemed that multiple reminders would be unacceptable. Completion and return of the questionnaire was taken to indicate consent to participate. The North West 2 and Lancaster University Research Ethics Committees approved the study.

Survey Instrument

Drawing on previous qualitative work regarding parental experiences of the consent process,26,32 a questionnaire was developed to explore parental decision making in relation to newborn blood spot screening. As part of this, and reported here, several sections evaluated components relating to the domains of informed consent. Parents who responded were taken to be competent to do so, and were asked to respond to a series of statements relating to (1) their perceived

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understanding of newborn screening, (2) the perceived availability of choice, and (3) their perceived ability to make a choice to accept or decline newborn blood spot screening.

**Perceived Understanding**

Given the limitations of asking parents to recall specific items of information, parents were asked to rate their perceived understanding of key screening elements. Questions were informed by previous research\(^3^2\) and asked parents to rate 9 statements regarding the motivation behind, and the process of, newborn screening, together with their understanding of the conditions for which newborn screening is conducted.

**Perceived Ability to Make a Choice**

To gauge whether respondents felt that they were capable of making a choice, 4 statements were presented. Statements reflected both situational factors (“I felt I had enough time to make a decision about the heel pricking”) as well as individual behavioral aspects (“I was too emotional to make a decision about the heel pricking”) although no formal assessment of competence was made.

**Perceived Availability of Choice**

Respondents’ perceptions regarding whether they had a choice to accept or decline newborn blood spot screening were assessed through responses to 4 statements. A final item asked parents to respond to the statement: “I feel I have made an informed choice.”

Responses to all statements were coded using a 5-point Likert-type scale from “Strongly Agree” to “Strongly Disagree.” Demographic information was also collected, together with household income, education, number of children, and age of respondent.

**Data Analysis**

Categorical responses were analyzed using simple descriptive statistics. Stepwise logistic regression analysis was performed to identify characteristics predicting perceptions of whether an individual had made an informed choice. The dependent variable was the response to the item “I feel I have made an informed choice,” using a recoded outcome (Strongly Agree/Agree vs Neither Agree or Disagree/Disagree/Strongly Disagree). All statements were initially included as independent variables within the model, together with 4 demographic predictors: income (≤£28,000 and >£28,000), education level (school/General Certificate of Secondary Education, college/Advanced Level General Certificate of Education, and additional education), age group (≤30 years and >30 years), and whether the parent was primiparous or multiparous (response options were combined because of small cell counts). Analysis was based on complete cases for all variables, and a backward stepwise process with Likelihood ratio testing. A significance level of \(P \leq .05\) was used as cutoff for retaining variables in the model. Data from completed questionnaires were entered and analyzed by using SPSS version 19.\(^3^6\)

**RESULTS**

**Parent Participation and Characteristics of Respondents**

Of the 500 parents approached to complete the questionnaire, 12 were subsequently excluded on the basis that the named individual no longer resided at that address. Of the remainder, a total of 154 questionnaires were returned giving a response rate of 32% (154/488). Only 3 responses were received through the online option, the remainder being returned by post. This response rate is similar to other survey research of parental experiences of newborn screening.\(^3^7\)–\(^3^9\)

Parents varied in terms of age, income, number of children, and educational level (Table 1). The sample was predominantly white (95.5%), aged between 31 and 40 (61%), and had either 1 or 2 children (79.9%). Respondents appeared to be more educated than the background population, with 84 of 154 (54.55%) having postcollege education compared with just 17.73% in the population from which the sample was drawn (based on 2001 census data).

**Perceived Understanding**

The majority of parents felt they understood the motivation behind newborn screening and the process...
The table 3 presents the parental responses to questions relating to their perceived ability to make a choice. Most parents provided responses indicating that they were able to make a choice whether to have their child screened. More than two-thirds of parents (68.1%) agreed to some degree that they had enough time to make a decision, and 83.1% felt they were able to make a decision about the heel prick. Parents, in general, did not feel that they were inhibited in making decisions, with only 10.6% feeling they were too tired to make a decision and 9.7% feeling they were too emotional.

Table 3: Parental Responses Regarding Perceived Ability to Make a Choice (N = 154), n (%)

<table>
<thead>
<tr>
<th>Statement</th>
<th>Strongly Agree</th>
<th>Agree</th>
<th>Neither Agree Nor Disagree</th>
<th>Disagree</th>
<th>Strongly Disagree</th>
<th>Missing</th>
</tr>
</thead>
<tbody>
<tr>
<td>I felt I had enough time to make a decision about the heel prick</td>
<td>27 (17.5)</td>
<td>78 (50.6)</td>
<td>23 (14.8)</td>
<td>21 (13.6)</td>
<td>4 (2.6)</td>
<td>1 (0.6)</td>
</tr>
<tr>
<td>I felt I was too tired to make a decision about the heel prick</td>
<td>4 (2.6)</td>
<td>13 (8.4)</td>
<td>24 (15.6)</td>
<td>85 (55.2)</td>
<td>26 (16.9)</td>
<td>2 (1.3)</td>
</tr>
<tr>
<td>I was too emotional to make a decision about the heel prick</td>
<td>5 (3.2)</td>
<td>10 (6.5)</td>
<td>20 (13.0)</td>
<td>88 (57.1)</td>
<td>30 (19.5)</td>
<td>1 (0.6)</td>
</tr>
<tr>
<td>I did not feel able to make a decision about the heel prick</td>
<td>6 (3.9)</td>
<td>8 (5.2)</td>
<td>10 (6.5)</td>
<td>90 (58.4)</td>
<td>38 (24.7)</td>
<td>2 (1.3)</td>
</tr>
</tbody>
</table>

The table 2 presents the responses to statements regarding perceived understanding of choice. Most respondents also provided responses indicating that they were able to make a decision whether to have their child screened. More than two-thirds of parents (68.1%) agreed to some degree that they had enough time to make a decision, and 83.1% felt they were able to make a decision about the heel prick. Parents, in general, did not feel that they were inhibited in making decisions, with only 10.6% feeling they were too tired to make a decision and 9.7% feeling they were too emotional.

Table 2: Responses to Statements Regarding Perceived Understanding (N = 154), n (%)

<table>
<thead>
<tr>
<th>Statement</th>
<th>Strongly Agree</th>
<th>Agree</th>
<th>Neither Agree Nor Disagree</th>
<th>Disagree</th>
<th>Strongly Disagree</th>
<th>Missing</th>
</tr>
</thead>
<tbody>
<tr>
<td>I feel I understand why the heel prick is done</td>
<td>46 (29.9)</td>
<td>82 (53.2)</td>
<td>12 (7.8)</td>
<td>9 (5.8)</td>
<td>3 (1.9)</td>
<td>2 (1.3)</td>
</tr>
<tr>
<td>I feel I understand why the test is done at the time it is</td>
<td>40 (26)</td>
<td>78 (50.6)</td>
<td>14 (9.1)</td>
<td>19 (12.3)</td>
<td>1 (0.6)</td>
<td>2 (1.3)</td>
</tr>
<tr>
<td>I feel I understand what the test results mean</td>
<td>28 (18.2)</td>
<td>86 (55.8)</td>
<td>23 (14.9)</td>
<td>14 (9.1)</td>
<td>0</td>
<td>3 (1.9)</td>
</tr>
<tr>
<td>I feel I understand how the test is done</td>
<td>38 (24.7)</td>
<td>93 (60.4)</td>
<td>10 (6.5)</td>
<td>10 (6.5)</td>
<td>0</td>
<td>3 (1.9)</td>
</tr>
<tr>
<td>I feel I understand when the test is done</td>
<td>34 (22.1)</td>
<td>95 (61.7)</td>
<td>13 (8.4)</td>
<td>7 (4.5)</td>
<td>1 (0.6)</td>
<td>4 (2.6)</td>
</tr>
<tr>
<td>I feel I understand when the results will be available</td>
<td>30 (19.5)</td>
<td>82 (53.2)</td>
<td>21 (13.6)</td>
<td>16 (10.4)</td>
<td>2 (1.3)</td>
<td>3 (1.9)</td>
</tr>
<tr>
<td>I feel I understand what the conditions are that the heel prick tests for</td>
<td>25 (16.2)</td>
<td>67 (43.5)</td>
<td>27 (17.5)</td>
<td>28 (18.2)</td>
<td>4 (2.6)</td>
<td>3 (1.9)</td>
</tr>
<tr>
<td>I feel I understand how the conditions will affect my child</td>
<td>23 (14.9)</td>
<td>65 (42.2)</td>
<td>22 (14.3)</td>
<td>35 (22.7)</td>
<td>7 (4.5)</td>
<td>2 (1.3)</td>
</tr>
<tr>
<td>I feel I understand how the conditions would be dealt with if found</td>
<td>15 (9.7)</td>
<td>38 (25.3)</td>
<td>38 (24.7)</td>
<td>48 (31.2)</td>
<td>11 (7.1)</td>
<td>3 (1.9)</td>
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The discussion section outlines the results of the study and the implications for future research. This study is the first to attempt to quantify parental experiences of the informed consent process in the context of newborn blood spot screening in the United Kingdom. The results of the survey indicate that the majority of respondents felt that they understood the purpose and process of newborn screening, but fewer felt they understood condition-related information. Most respondents also...
indicated that they felt able to make a decision. However, there was much less agreement regarding the perceived availability of choice, with the majority of parents indicating that they felt the heel prick was expected, and almost a third indicating that they did not feel they had a choice to decline the test. Despite this, more than two-thirds of respondents indicated that they had made an informed choice, although a substantial minority felt they had not. These findings should be interpreted in light of the limitations of the study. The response rate of 32%, although comparable to other survey research relating to parental experiences of newborn screening that have seen response rates as low as 23%,37 28.6%,38 and 32.5%,39 is relatively low. The responses here are also limited to those parents who accepted screening, and it may be that those parents who decline screening may have different experiences, particularly in relation to the perceived ability to decline screening. Additional research specifically considering those parents who decline screening would be of great benefit. Despite these limitations, our results provide illuminating insight into the perceived understanding of newborn screening and the role that the presentation of screening has on perceptions of informed choice.

Previous studies have suggested that parental knowledge and understanding of screening may be low.4 25,28,40 Although knowledge of the names of conditions or their major cause has tended to be relatively low, knowledge of condition-specific information has generally found to be better.37,41,42 Research in the Netherlands, for example, found that knowledge of recurrence risk was good within a small cohort of patients identified as having cystic fibrosis, with 94% knowing their recurrence risk.43 Our finding that parents indicated a greater understanding of the motivation and process of screening, as opposed to the screened for conditions, stands in contrast to this. This likely reflects the more general population of our sample as opposed to the samples of Dankert-Roelse and colleagues,43 as well as others37,42 who have considered subpopulations, often after a diagnosis.

Our results suggest that when there is no contraindicating information about the health of the child, then condition-specific understanding may be low, a result that is consistent with recent work in the United Kingdom in which parents were found to have low levels of knowledge about the conditions included and what they would mean for their child.24

Our findings also indicate several service provision factors that may improve the consent process. Respondents were more likely to indicate that they had made an informed choice if they felt they understood the reasons for screening, if they felt that the test was presented as an optional test (ie, they had the opportunity to choose), and if they felt they had enough time to make a decision.

The need for time to make a decision is congruent with results from numerous studies regarding the provision of information to parents and in which a consistent finding is that parents want information in the prenatal period.10,29,33,44 In the United Kingdom, for example, guidance indicates that information should be given in the third trimester of pregnancy. However, it may also be given postnatally, so long as this is up to 24 hours before the heel prick.15 Ensuring that information about newborn screening is given in the third trimester may not only mean that parents have time to look at this information but may facilitate informed decision-making by allowing more time in which to make a decision. Indeed, the finding that multiparous parents are more likely to feel that they have made an informed choice is compatible with having time to make decisions. Parents for whom this is their second or third round of screening have already been through the decision-making process, and consequently, it may be easier for them to make decisions and feel that they have made an informed choice because this decision is informed by
their previous experience. This supposition gains some support from research into decision-making for prenatal testing in which personal experience was a significant factor.45,46

The present results also lend support to previous work that has found that, despite an explicit mandate for informed choice, parents may experience newborn blood spot screening as a routine procedure.25,27,47 Such a finding appears not to be limited to the United Kingdom and has been found internationally.33,48 Our results indicate that an important aspect of this is the way that screening is perceived to be presented; almost 80% of respondents indicated that they felt it was expected that their child have the heel prick, and less than half felt that it was presented as an optional test or that they could decline. Research by Parsons et al.,47 which sought to evaluate an intervention that emphasized an optional screen for Duchenne muscular dystrophy, found that parents who received the intervention were more likely to feel that the midwife had given them a choice and were significantly more satisfied than those who did not receive a separate invite. Consequently, the presentation of the information, and choice, appears to play a crucial role in parental experiences of decision-making for newborn blood spot screening.

Finally, our finding that those who feel that they understand the motivation behind screening are more likely to feel that they have made an informed choice is important from an educational perspective. If the aim of these information materials is to promote informed choice, then our results indicate that important content should be the reason for newborn screening. Current information, in the form of the “Screening Tests for You and Your Baby” booklet, clearly indicates that an important motivation for screening is to identify babies who have rare but serious conditions and that early identification is beneficial to the health of the child. However, studies have suggested that the recall of having this written information is somewhat limited.44 The same research has also indicated that information provision by the healthcare professional (primarily the midwife) during the pre- and postnatal visits was seen to be beneficial because this was time that had been already set aside and did not require the active seeking of information. It is not clear what impact information (written or verbal) is having on parental decisions regarding newborn blood spot screening. Although studies have suggested that current written information about screening programs do not promote informed choice,48 and despite the resources spent on the development of information materials,50 there is a paucity of work in terms of evaluating their quality or their impact on parents’ decisions. A clear understanding of the impact that information content and/or delivery has on parental decisions and decision-making is essential to optimize resources and inform strategy for information provision.

Our results suggest that by attending to the content of information, principally the motivation for screening, and presenting this in a timely manner, it may be possible to facilitate informed decision-making by parents and therefore promote an informed choice and consent for newborn blood spot screening.

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REFERENCES

36. IBM. SPSS, version 19. Chicago, IL: SPSS Statistics; 2012
42. Lewis S, Curnow L, Ross M, Massie J. Parental attitudes to the identification of their infants as carriers of cystic fibrosis by newborn screening. J Paediatr Child Health. 2006;42(9):533–537
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