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ARTICLE

Consent for newborn screening: parents' and health-care professionals' experiences of consent in practice

Holly Etchegary*[,1], Stuart G Nicholls[2,3], Laure Tessier[4], Charlene Simmonds[5], Beth K Potter[2], Jamie C Brehaut[6], Daryl Pullman[7], Robyn Hayeems[8], Sari Zelenietz[4], Monica Lamoureux[4], Jennifer Milburn[4], Lesley Turner[9], Pranesh Chakraborty[4,10] and Brenda Wilson[2]

Consent processes for newborn bloodspot screening (NBS) are variable, with a lack of descriptive research that depicts how the offer of NBS is made to parents. We explored the experience, in practice, of consent for NBS. Semistructured interviews in two Canadian provinces were held with: (1) parents of children offered NBS ($n = 32$); and (2) health-care professionals involved in the NBS process ($n = 19$). Data on recollections of NBS, including consent processes, were utilized to identify emerging themes using the method of constant comparison. Three themes were relevant to NBS consent: (1) The 'offer' of NBS; (2) content and timing of information provision; and (3) the importance of parental experiences for consent decisions. Recollections of consent for NBS were similar between jurisdictions. Excepting midwives and their patients, NBS was viewed as a routine part of giving birth, with little evidence of an informed consent process. Although most parents were satisfied, all respondents suggested information about NBS be provided long before the birth. Accounts of parents who declined screening highlight the influence of parental experiences with the heel prick process in screening decisions. Findings further our understanding of consent in practice and highlight areas for improvement in parent–provider interactions.
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INTRODUCTION

Newborn bloodspot screening (NBS) is one of the oldest population-based screening programs.[1–4] It involves taking a small amount of blood from a baby's heel shortly after birth and testing it for a number of serious conditions. The severity and treatability of paradigm conditions such as phenylketonuria and congenital hypothyroidism demanded early detection, garnering international support for NBS.[4–6]

Tandem mass spectrometry and next-generation sequencing allow the expansion of NBS panels to include conditions for which the natural history of the disorders is less clear, with debate regarding the balance of clinical risks and benefits to the affected infant.[5–9] A wider range of potential benefits for families have been proposed, including guidance for family planning, earlier diagnosis and avoidance of the 'diagnostic odyssey'.[8–10]

NBS programs differ considerably with respect to parental educational materials[6,11–13] and approaches to parental consent for screening.[6-8,14,15] In Canada, screening is considered routine and proceeds unless parents explicitly object – an opt-out approach.[6,16–18] In contrast, some US states have legislation requiring explicit consent, whereas others institute an opt-out approach.[19–21] Other jurisdictions, such as the United Kingdom, offer NBS strictly on an informed choice basis.[22] Disparities between jurisdictions with regard to the need for explicit consent, ongoing discussion regarding the provision of information about NBS before sample collection and what level of understanding is necessary to ensure informed consent have renewed research interest in informed consent practices.[23,24]

To date, the literature concerning consent and newborn screening has generally focused on attitudes toward consent policy.[7,25–29] Although this is revealing, occasionally showing conflicting attitudes toward the need for consent,[6,30,31] there is a lack of descriptive research that depicts how the offer of NBS is made to parents, making it difficult to determine the experience of consent in practice. Understanding the experiences of those actually involved in the consent process may provide valuable descriptive information for NBS programs and providers. This study explores parent and health-care provider (HCP) experiences of NBS consent practices in two Canadian provinces. The findings further our understanding of consent approaches and highlight potential areas for improvement in parent–provider interactions.

MATERIALS AND METHODS

This qualitative interview study was approved by health research ethics boards in both Ontario (ON) and Newfoundland and Labrador (NL). The full study protocol is available elsewhere[32] and the methods are summarized below.

[1]Clinical Epidemiology Unit, Faculty of Medicine, Memorial University, St John's, Newfoundland and Labrador, Canada; [2]School of Epidemiology, Public Health and Preventive Medicine, University of Ottawa, Ottawa, Ontario, Canada; [3]Children's Hospital of Eastern Ontario (CHEO) Research Institute, Ottawa, Ontario, Canada; [4]Newborn Screening Ontario, Children's Hospital of Eastern Ontario, Ottawa, Ontario, Canada; [5]Health Research Unit, Faculty of Medicine, Memorial University, St John's, Newfoundland and Labrador, Canada; [6]Ottawa Hospital Research Institute, Ottawa, Ontario, Canada; [7]Community Health and Humanities, Faculty of Medicine, Memorial University, St John's, Newfoundland and Labrador, Canada; [8]Institute of Health Policy Management and Evaluation, University of Toronto; Program in Child Health Evaluative Sciences, Hospital for Sick Children Research Institute, Toronto, ON, Canada; [9]Provincial Medical Genetics Program, Eastern Health, St John's, Newfoundland and Labrador, Canada; [10]Department of Pediatrics, Faculty of Medicine, University of Ottawa, Ottawa, Ontario, Canada
*Correspondence: Dr H Etchegary, Clinical Epidemiology Unit, Faculty of Medicine, Memorial University, Craig L Dobbin Centre for Genetics, Room 4M210, St John's, Newfoundland and Labrador A1B 3V6, Canada. Tel: +1 709 864 6605; Fax: +1 709 864 6529; E-mail: holly.etchegary@med.mun.ca
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Research sites

Interviews were conducted concurrently in ON and NL. The largest program in Canada, Newborn Screening Ontario (NSO), screens for 29 disorders in over 140 000 samples annually. Of these, ~1300 generate screen positive results, with roughly 200 confirmed positive at diagnostic testing. Conversely, NL screens for 7 core conditions, in ~4500 live births annually; there are 40–50 screen positive results and ~2 confirmed at diagnosis. The number of confirmed, true positive screening tests for both programs varies year to year, but ranges from 3 to 5% in NL and from 10 to 15% in ON. NBS is declined for 1–2 infants per year in NL, and <100 in ON.

Sampling and recruitment

Parents. Purposive sampling[33,34] based on screening result was used to identify parents of newborns in four groups (negative, false positive, true positive, declined) to ensure a broad range of experiences with NBS. Parents were identified through provincial screening program records. Eligibility criteria were age ≥18 years, having a child eligible for screening in the preceding year and conversant in English or French. Parents were excluded if their child was known to have died or was very ill, had been adopted or was under the care of Children's Aid. For screen negative infants, where information was available in the NBS programs' database, we excluded parents whose child was transfusion positive or born before 35 weeks of gestational age; a research ethics board suggested the latter outcomes could indicate poor health and a study invitation might be distressing.

A geneticist or genetic counselor reviewed all records to determine eligibility. Potential participants were sent an invitation letter, information sheet, consent form and return slip. Because of the very low numbers at the NL site, and in order to maximize recruitment, parents of newborns with a true or false positive result, and those who had declined screening, were called directly by the geneticist who had provided care before the mailed information. He or she advised about the study and the subsequent mailed information, and answered any questions. When mailed recruitment methods failed to recruit parents of screen negative infants at the NL site, they received a follow-up phone call 2 weeks after the mailed information. At the ON site, parents who had declined NBS were approached through the HCP responsible for the care of their child to reduce any perception of coercion on the part of the screening program. These HCPs were provided with a study information package and asked to forward these to the identified parents and discuss the study with them.

Health-care professionals. HCPs who submitted blood spot samples to the provincial screening program, and those providing counseling or education to parents regarding NBS, were eligible. Purposive sampling was used to recruit HCPs in different professional groups, including midwives, obstetricians, pediatricians, maternal/newborn nurses, family physicians and genetics professionals. They were identified through information contained in screening reports, specialty/professional networks and the research team's networks. HCPs were advised about the study by phone or e-mail and sent a study invitation letter, consent form and return slip.

Established methods were used to determine a sample size likely to be sufficient to ensure data saturation (the point at which no new information is gained from interviews).[33,34]

The interviews

Interviews were conducted in person or by telephone, ranged from 30 to 60 min, were audio-recorded and transcribed verbatim. They covered two key areas: (1) recollections of experiences with NBS; and (2) attitudes toward various models of consent for NBS. This paper addresses the first area. For parents, the focus was on the offer of NBS, information they received and their satisfaction with the process. HCPs were asked to recount their experiences (if any) with offering NBS and their perception of parents' understanding of NBS.

Data analysis

Qualitative description[33] was used to summarize the data pertaining to experiences of consent for NBS. This form of naturalistic inquiry makes no theoretical assumptions about the data. Instead, it presents the data in the language of participants, without necessarily aiming to interpret the data in more theoretical ways. The end result is a comprehensive summary of the event in question.[33]

Transcripts were read and re-read several times by one investigator (HE). Interview data were then isolated and organized around interview topics (eg, experiences of NBS, attitudes towards consent, etc). Data pertaining only to personal experiences of NBS were utilized to identify and index emerging categories and themes for the current paper. Two investigators (HE and SGN) then separately read and re-read the isolated data, and used the method of constant comparison to inductively subcode the data relevant to NBS.[33,34] Essentially, data were compared between and within transcripts and the two study samples to establish analytical categories and themes,[33,34] with a constant shifting back and forth between and within transcripts to compare the perceptions and experiences of participants. When both investigators had completed their separate analyses, they discussed categories and themes. Agreement was very high, and the analysis was then presented to the broader research team for discussion.

Table 1 Participant demographics

Item	N *(%)*
Parents	32
Screen negative	13 (41%)
True positive	9 (28%)
False positive	7 (22%)
Declined screening	3 (9%)
Ontario	19 (59%)
Newfoundland and Labrador	13 (41%)
Health-care professionals	19
Ontario	15 (79%)
Newfoundland and Labrador	4 (21%)
Interview total	51

Because of small cell counts, some groups have been collapsed in order to mitigate concerns regarding identification.

RESULTS

Participants

In total, 51 interviews were conducted with parents (32) and HCPs (19) (Table 1). All parental interviews except two were with mothers. Parents reported screening being conducted by both clinical and laboratory personnel. This range of personnel was reflected in our HCP sample that comprised midwives, nurses, nurse educators, laboratory professionals or managers, pediatricians and a genetic counselor.

Although parents reported a range of experiences, largely based on variation in screening outcome, the narratives of both parent and professional groups were consistent. Experiences of consent for NBS in practice were encompassed by the following themes: (1) the 'offer' of NBS; (2) parental information about NBS: needs and timing; and (3) how parental experiences influence consent.

The 'offer' of NBS. Two different accounts of NBS as a choice were offered by participants. In a minority of parental accounts – largely those under the care of midwives – NBS was recalled as a clear choice. Among the HCP interviews, only midwives were consistent in describing a conscious informed consent process.

> The midwife explained to me that the newborn screening would help to screen the child for any potential diseases. … It was definitely presented to me as a choice, one hundred percent (parent 9, declined screening).

They were very concerned about informed consent … She did her best to provide as much information as she could to help us make the most informed choice that we could (parent 10, true positive result).

I would never dream of just providing a pamphlet. We certainly give them the written information, and we'll go over that with them, but mostly, we do it verbally. I certainly don't expect anybody to just let me do procedures on their baby they don't understand or agree to (HCP 10, midwife).

In contrast, the dominant account by both parents and HCPs suggested that NBS was neither offered nor experienced as a choice:

Moms are not asked any consent for the newborn screen … it's just considered a routine procedure (HCP 16, pediatrician).

I don't know if it was a nurse… It was post-partum, it was a bit blurry … I guess I signed the consent?... I don't really remember when they even did the heel prick. I can't believe it (parent 25, screen negative result).

The process was highly routinized, sometimes even described as being presented as compulsory:

Most of the nurses just tell the parents that this is a blood test that every baby in [province] gets to rule out different anomalies. Nobody goes into much detail (HCP 8, nurse).

I proceed to go to the room where the mother and baby are, confirm all the information, and then we draw the blood. There's never been permission for it (HCP 5, laboratory professional).

So she said, 'Well it's the law, you have to get it done' … I just said, 'I don't care, he's screaming and I don't want it done, and as far as I know from my midwife, it was voluntary.' … and then I think the nurses came in said, 'Oh it's hospital policy that you have to get it done within this hospital before leaving,' and we said 'No' (parent 32, declined screening).

Parents expressed a range of attitudes toward the importance of explicitly seeking consent:

I kind of find it disgusting [routine blood draw] because it doesn't give any parent a right … it doesn't take spiritual beliefs or faith into consideration. It's very presumptuous that they know what's right (parent 9, declined screening).

It didn't really strike me as anything that was a big deal, just part of the birthing process (parent 2, true positive result).

It wasn't an offer, but I was fine with how it was done. I would want to know if anything was wrong (parent 26, screen negative result).

Are you allowed to say no to newborn screening? I assumed that it was just something that everyone did. And upon reflection, I definitely would have continued to say yes (parent 13, false positive result).

Parental information about NBS: needs and timing. The identification by HCPs of NBS as a routinized activity was in no way accompanied by a sense that parents did not need to know what was happening to their child. Recognition of the diverse information needs of parents was expressed across multiple interviews:

This day and age, people are wanting to be more aware of what's being done and for what reasons. I think they need to understand the tests, why they're being done and the issues should the baby be positive (HCP 7, nurse).

They need to be informed not only what the testing is doing, but what to expect when the person comes to do it. … Patients seem to want to be more informed … more involved in their own care (HCP 6, laboratory professional).

There was a lot of questioning around the information sheet we give to the parents. It doesn't really explain as much as it could …What does it mean to them and what does it mean to their child? (HCP 3, laboratory professional).

The accounts of parents and HCPs were very consistent in agreeing that information about NBS should be provided to parents before being admitted to hospital for the birth, preferably by the HCP providing care throughout pregnancy:

Your family physician should discuss it with you later in pregnancy. … it would be a good opportunity to discuss it and ask questions. You should be given something [on paper] that you can refer to later (parent 26, screen negative result).

I may have been told about it while I was in labor, but I have no recollection whatsoever. Even before, by my family doctor or OB, not while in labor (parent 19, true positive result).

I'm not sure she [Mom] wants to hear about newborn screening at 24 hours. So, way before the baby is delivered (HCP 17, pediatrician).

Whether it starts at the actual prenatal visits… – this is the test that will be done when your baby is born – and continues through labor and delivery … then they should already be well-prepared for the fact that we're doing it (HCP 8, nurse).

Midwives' accounts were again distinct from the other HCPs. They described a typical practice of discussing NBS with parents long before labor and delivery and linked this with promoting informed parental decision making:

In the prenatal period, we talk about all the reasons why we would recommend the newborn screen, but we also do offer them the choice to opt out … we have a pamphlet that goes home with them... we want families to have a chance to talk about it and make sure it's right for them (HCP 12, midwife).

The narratives of parents whose child received a screen positive result suggested that information not only supported an initial consent decision, but also served to reduce distress and confusion. Receipt of a screen positive result was often the point at which accurate understanding became highly salient.

I was called at home … it kind of threw me off because I was, 'okay, newborn screening? I'm not really sure what she's talking about.' She told me he tested positive for this and needed to have repeat blood work and urine and of course, then I started to freak out, so I found that very stressful (parent 24, false positive result).

I think if I had more information – just because you get a call, doesn't mean a hundred percent you're going to have something wrong. I was fully in panic mode for a few days until we got in there … and they called the same day with the test results and everything, so that eased my mind fairly quickly (parent 7, false positive result).

As a new parent, it's a very stressful thing to get that phone call, and I just felt there was a lack of communication when we got the call saying she had tested positive. They asked us to rush into the hospital that day. … I felt like there wasn't very much information explaining what could be wrong and we had to wait about three months for test results (parent 8, true positive result).

How experiences influence consent: the heel prick 'trauma'. Parents' decisions were also shaped by their experience of the blood draw, particularly those few parents who declined screening ($n=3$). The perception that their child was in great pain during the heel prick seemed key to their subsequent decline.

We had a terrible experience [NBS with first child]. I think this was just torturing the baby. With the second baby, I personally wouldn't have done it the second time, but I didn't know it was not mandatory... now, the third baby, the midwife told me that it is actually not mandatory, so then I said, 'well, then I won't even have it.' I found the decision very easy (parent 16, declined screening).

The lab technician came to do it, which probably took her about 7 minutes of squeezing his foot and he was screaming his head off … it was just so awkward … another 5 minutes to get one circle filled for the newborn screening. … I said, 'you need to get out of here. I don't care enough about newborn screening for you to be doing that and him screaming.' … I would feel better about doing a consent form or where you have to say yes (parent 32, declined screening).

DISCUSSION

In this study, we presented descriptive qualitative data on perceptions of how two Canadian NBS programs appear to work in practice, from both provider and parent perspectives. There were few discernable differences in accounts across study sites, perhaps unsurprising as NBS proceeds as a standard of care in Canada, even if not legally mandated.[17,29]

Results are consistent with a growing literature revealing very supportive parental views of NBS.[27,29,35–36] It is important to appreciate that the majority of parents in NBS programs are 'satisfied customers':[37] most will receive screen negative results, and in the case of acceptance of screening, their understanding is not put to the test. It highlights the importance, in research and quality assurance studies, of seeking out those participants who are most likely to provide a discordant view, however infrequent this is. By definition, a screening program that targets rare disorders will have an overwhelming majority of parents of healthy infants. With the passage of time, these parents are probably the most likely to forgive or diminish concerns about principles or process. Deliberately seeking out participants with divergent views, such as those under the care of a midwife or those who have declined the screening program, may be key to identifying potential areas for improvement as they may be less forgiving of perceived suboptimal care or consent processes. In an effort to seek divergent perspectives, we actively recruited parents who declined newborn screening and parents of children who received false positive results. In smaller programs where the absolute number of such cases each year is small, there may be a tendency by clinicians and research ethics boards to wish to particularly protect these parents, in terms of privacy, potential for distress or coercion to participate. As researchers, we must take these concerns seriously. However, we have demonstrated that it is possible to conduct such research in a sensitive and responsible way, and that the data may be highly illuminating. For example, we gained important insights about the reasons that some parents decline that would not have been evident from interviews with only those who accepted screening. We encourage other groups to be strategic about including these parents in future studies.

The picture that emerged from this study was that NBS happened as a standard part of the birthing process, with limited conversation about it before the blood spot was taken. The norm among HCPs was not to seek consent from parents, the exception being midwives as a group: their own accounts, and those of parents, were consistent about their commitment to informed decision making. Our findings are congruent with previous research that noted that the issue of consent is complex, with little consensus on the amount of parental involvement required for screening.[6,30,31] Other studies reported that providers who viewed obtaining consent as relatively easy (eg, midwives) appeared to favor an informed consent approach, in contrast to many providers who saw the process as time consuming and impractical (eg, pediatricians).[6]

Meaningful parental understanding about NBS is a prerequisite for informed consent, but the importance of education is relevant even where screening is mandated. Several studies have found that having adequate information improved parents' experiences of positive screens.[31,35,37] In our study, such parents recounted confusion and distress, with some having no memory that their child had even been screened. Providing key information well before delivery could help alleviate later distress by preparing parents for the possibility of positive screening results, and reinforcing the difference between screening and diagnosis. Thus, although there is still room for discussion about the specific content of parental education, whose responsibility it is to offer it and effective ways of providing it,[9,17,36] our findings add support to the importance of efforts to ensure parents go into the birthing process already knowing about NBS.[27] These results are not new, but they do raise interesting questions for researchers and those responsible for NBS programs. For example, findings challenge us to explore what kind of information seeking parents engage in before labor. Is there any onus on parents to engage in information seeking before the birthing process? Other interesting questions relate to why one group of HCPs view informed consent as less onerous than others or to study actual HCP–parent appointments with a view to explore why (or if) information on NBS is not routinely offered in prebirth appointments. These research foci could help inform practical processes for parental information provision and consent.

Our results also indicate how parents' decisions to decline screening may not be based solely on a principled aversion to screening, but rather on prior birth experiences or of the heel prick process. Two parents in this study declined on the basis of their experiences of perceived distress caused to their infant during the heel prick. The fact that these decisions were not principled objections to NBS, but were based on previous screening experiences, suggests that parental consent is not only a question of parental education and values, but also a quality issue with respect to blood draw procedures. It is important to ensure that blood draw practices are informed by the current literature that point to the analgesic benefits of skin-to-skin contact,[38] breastfeeding[22,39] as well as best practices regarding the temperature of the heel when conducting blood draws.[22,40]

The experiences of parents who declined screening following the emotionally traumatic experiences of the blood draw are illustrative of the contextual nature of consent for newborn screening, as are comments reflecting the way screening is presented. The role of the situational context of the consent process – what actually happens in practice – has largely been absent from discussions of consent for NBS. Our findings suggest a need to include consideration of the way in which consent processes are managed within newborn screening, but also point to the important role of professional education and training with respect to the consent process.

Finally, we concur with others[29] who suggest the need for caution around the potential consequences for parents as traditional screening methods are replaced by new genomic technologies such as next-generation sequencing. These methods may increase the number of positive newborn screens (true or false), and our data suggest that positive screening results have the potential to increase distress if parents are poorly educated or lack awareness of testing. Unless efforts are made to increase understanding and awareness among parents in screening programs, parental distress associated with NBS will increase.

CONCLUSIONS AND RECOMMENDATIONS

This study confirms previous research that consistently reveals largely positive views of NBS. The perception of NBS as a standard part of the birthing process with little discussion among HCPs and parents before the blood spot collection is also well reported. However, the study adds new information by its inclusion of midwives and parents under

a midwife's care, as well as decliners of NBS. Although small in number, these accounts provide discordant views from the majority attitude about NBS. Further study of these accounts could help illuminate aspects of midwifery that facilitate awareness and informed decision-making among parents, as well as promote understanding of decliners of NBS programs. These latter accounts encourage researchers and NBS programs to think about consent in ways other than a rational, informed process. Rather, decisions about NBS may be based on experiential knowledge, rather than technical knowledge of the tests' risks and benefits. Study findings are also noteworthy in that they revealed some parents believed NBS was mandatory. This belief does not correspond with choice about NBS in Canada, and better discussion among HCPs and parents is needed if we are to facilitate parent choice in this area. Finally, our data reveal parental distress following a positive screen result (true or false). This suggests that efforts are needed to promote parental understanding and awareness of NBS before the blood spot collection. Implementation research is needed to provide evidence-based recommendations regarding the most effective way of providing parental education about NBS.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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