Parental consent for newborn screening: a discrete choice experiment

by © Yifu Liao

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Abstract

Background: Parental consent is very commonly assumed for newborn bloodspot screening (NBS) in most Canadian provincial screening programs. This falls short of usual norms, and evidence suggests that some parents would prefer an explicit process. This study was designed to inform improvements in NBS consent processes.

Objectives: (1) To examine parents' past experiences with, and attitudes towards, NBS consent processes in Canada. (2) To quantify parents' preferences towards specific attributes of the NBS consent process, and identify characteristics of subgroups with different preference patterns.

Method: A cross-sectional survey that included a discrete choice experiment (DCE) was conducted to capture information on participants' past experiences with and preferences for NBS consent processes. DCE data were analyzed using conditional logit and latent class (LC) regression models.

Results: The sample comprised 715 participants. As an overall group, respondents preferred to have NBS information provided late in pregnancy, for consent not to be assumed by providers, and for the consent decision to always be recorded. Three classes of participants with different underlying preference patterns were identified in the sample.

Conclusion: If NBS programs wish to better meet parents' preferenes, the results indicate specific aspects of the consent process that could be targeted for further examination..

General Summary

When being offered medical or preventive treatments, informed consent by patients is a standard expectation. However, in practice, many screening tests for newborn infants are often carried out on an assumption that the parents consent, without any formal process to document this. Evidence suggests that this goes against the expectations of some parents. This study examined parents' preferences for aspects of the consent process for newborn screening in more depth. Using data from a survey of parents of young children, we examined past experience, attitudes, and preferences on this topic. The overall results suggested that they preferred to receive informational material about screening in late pregnancy, for their consent not to be assumed, and that their decision should always be formally recorded. Within the overall sample, however, we identified one group who prioritized the formality of the consent process, and one that prioritized information and time for decision making.

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DMCI: Decision-Making Control Instrument

HIOS: Health Information Orientation Scale

LC: Latent Class

NBS: Newborn Bloodspot Screening

OR: Odds Ratio

SD: Standard Diviation

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CHAPTER 1 Introduction

The focus of this thesis is the consent of parents for screening of newborn infants for a range of rare conditions. Newborn bloodspot screening (NBS), in particular, is offered in every Canadian province, to check for a number of rare conditions that do not cause symptoms until later in infancy or childhood (Hayeems et al., 2013). Quite often it is assumed that parents have given their consent to this screening. However, in recent years as the numbers of conditions that can be screened for has expanded, and it is not clear whether or not parents understand and consent to having their child screened (Etchegary et al., 2016; Nicholls et al., 2019). This thesis aims to examine the issue of informed consent for newborn screening.

A patient's right to accept or decline healthcare interventions is a fundamental principle of medical practice. There are four basic principles commonly invoked in health ethics: respect for autonomy, beneficence, non-maleficence, and justice (Beauchamp & Childress, 2001; Gillon, 1994). Consent is an important aspect in these principles in health ethics. A reasonable patient will consent when they perceive that the benefits of participation outweigh potential harms. Obtaining a valid consent demonstrates respect for patient autonomy and justice (respect for people's rights (Gillon, 1994)).

Consent for NBS is complicated since parents have to make a decision on behalf of newborn infants based on what is good for them. Currently, based on observations made by parents and healthcare providers, consent for NBS appears to be assumed, rather than explicitly sought, in the majority of Canadian provinces (Potter et al., 2015; Etchengary et al., 2016; Nicholls et al., 2019).

The principle of consent has been practiced during surgical procedural since the 18th century (Jones, McCullough, and Richman, 2005). In the beginning, the simple consent model

was commonly used. Simple consent involving asking one question (Jones, McCullough, and Richman, 2005): "Did the patient agree to be treated?" A landmark opinion by Judge Benjamin Cardozo in a 1914 U.S. court case established that every competent human has the right to decide what should happen to his or her body (Jones, McCullough, and Richman, 2005). The same principle has been upheld in Canadian case law (Pullman, 2001).

Obtaining a signature on an authorization form may satisfy a legal requirement, but it does not necessarily satisfy the ethical requirements for informed consent (Jones, McCullough, and Richman, 2005; Katz and Webb, 2016). Many people sign legal documents without fully reading or understanding them (Wogalter, Howe, Sifuentes, and Luginbuhl, 1999). In an attempt to satisfy perceived legal standard of informed consent, consent documents are often excessively technical and may contain too much information (Pullman, 2001). Technicality, lengthiness, and illegibility were frequently reported reasons for not fully understanding legal documents (Wogalter, Howe, Sifuentes, and Luginbuhl, 1999). Informed consent should be about the dialogue between the physician and the patient in deciding the course of action in managing their health condition (Katz and Webb, 2016). How can we satisfy the ethical requirement of consent if obtaining a signature on an authorization form might satisfy a legal requirement, but nevertheless fails to satisfy the ethical requirement?

According to Kinnersely et al. (2013), a valid informed consent is given when the patient has done the following:

- Understood the information provided;
- Retained enough information to make the decision;
- Weighed the information in the decision-making process; and
- Communicated their decision to their healthcare professionals.

It is important for the patient to have enough information and the compentency to assess the information and make the decision. Thus, in order to achieve a valid informed consent, first, the patient should be provided enough information in a language that they understand (Katz and Webb, 2016). The content of the information should include the nature of their condition, proposed treatment, potential alternatives, and benefits and risks of each alternative. First, the capacity of the patient to make a decision must be assessed. Assuming the patient is competent, their understanding of the information provided should be assessed (Katz and Webb 2016). Finally, the patient's autonomy in making the treatment decision should be assured (Katz and Webb, 2016). The patient should be able to choose among all alternatives without being influenced or manipulated.

The two major elements of an informed decision are that the decision is (a) based on adequate knowledge and understanding by the decision-maker, and that it is (b) consistent with their values and preferences. In Canada, physicians are legally and ethically obligated to provide what a reasonable patient would want to know. Failure to do so would not make the consent invalid but it may constitute negligence, a breach of physicians' duty of care (Tigerstrom, 2001; Robertson, 1991). To determine whether enough information is provided, two standards emerged from the 1950's to the 1970's. These standards are the "professional practice" standard: what an experienced physician in the community would tell about the patient's condition; and the "reasonable person" standard: what a reasonable person would want to know about their condition in order to make the decision. The professional practice standard emerged before patient's autonomy became an important concern in health ethics (Pullman, 2001), and it was rejected in 1972 due to the growing skepticism of the integrity a solely physician-based standard (Jones, McCullough, and Richman, 2005). The reasonable person standard was established after

the court case of *Halushka v. University of Saskatchewan* (1965). The court case of *Reibl v. Huges* (1980) strengthened and modified the reasonable person standard which became the "modified objective standard" (Pullman, 2001). According to the court proceedings, Reibl sued Hughes for negligence after suffered a massive stroke that caused paralysis on the right side of his body immediately after a surgery performed by Hughes. Reibl claimed that Hughes did not inform him about the possibility of paralysis shortly after the surgery and that the surgery could be postponed. Instead, Hughes stressed that the chance of paralysis would have been greater if Reibl had not undergone surgery. One crucial factor in this case was that Reibl was less than two years away from being eligible for a pension from his employer, so was anxious to continue working (Tigerstrom, 2001). The paralysis caused by the stroke after the surgery had interfered Reibl's ability to work. Hence, in the modified objective standard, we always ask "What would a reasonable person *in the patient's position* would want to know?" If a reasonable person in the patient's position would act differently when certain information is disclosed, then not disclosing that may constitute negligence.

1.1 Background to Screening in Healthcare

The balance of harms and benefits may be different for a preventive health intervention than for a therapeutic intervention. Over 50 years ago, Wilson and Jungner (1968) published their criteria for screening amidst controversy about early monitoring and detection of diseases (Andermann, Blancquaert, Beauchamp, and Déry, 2008; Becker et al., 2011). The goal of these screening criteria (shown in Box 1) was to provide a framework for assembling evidence on a proposed screening program, in order to ensure a holistic assessment of benefits and harms, and to take a system perspective on screening compared with other demands on resources. These criteria focus attention on the capacity to detect the condition with accuracy, the effectiveness of

treatment available for the condition, and the health system implications of offering screening in a fair and sustainable way.

Box 1: Wilson & Jungner Criteria for Screening (Wilson & Jungner 1968):

- Knowledge of disease
  - Condition must be an important health problem
  - Recognizable latent or early symptomatic stage
  - Natural course of condition (including development from latent to declared disease) should be adequately understood
- Knowledge of test
  - Suitable test or examination
  - Test acceptable to the population
  - Case finding should be a continuous process and not 'once and for all' project
- Treatment for disease
  - Accepted treatment for patients with recognized disease
  - Facilities for diagnosis and treatment available
  - Agreed policy concerning whom to treat as patients
- Cost considerations
  - Costs of case finding (including diagnosis and treatment of patients diagnosed) economically balanced in relation to possible expenditures on medical care as a whole.

1.2 Newborn Bloodspot Screening (NBS)

As technology has improved, the number of diseases for which the pre-clinical or pre-pathological biomarkers is proposed has increased (Andermann, Blancquaert, Beauchamp, and Déry, 2008). Screening for a genetically inherited disease (phenylketonuria, PKU) in newborn infants was implemented in large scale in the 1960s (Grosse et al., 2006), thanks to the development of a test based on Guthrie's bacterial inhibition assay (the "Guthrie test"). PKU is a rare genetically inherited inborn error of metabolism that can cause neurological damage if left untreated (Carpenter et al., 2018). Early detection of PKU in newborn children allows parents to implement phenylalanine-restricted diets in order to prevent permanent brain damage (Berry et al., 2013).

The Guthrie test technology was superseded in the 1990s and early 2000s by tandem mass spectrometry (MS:MS), which permitted the rapid detection and measurement of a wide range of metabolites using a single bloodspot sample. In Canada, NBS panels cover 5 to 38 conditions depending on jurisdiction (Hayeems et al., 2013). PKU and congenital hypothyroidism are screened across all provinces, mandatory in one of them. Treatments for PKU and congenital hypothyroidism are available that alter the outcome, such that affected children are likely to live a more normal life. The disease group of which PKU is a member – the inborn errors of metabolism – includes a large number of conditions. These make up by far the most common conditions screened for in NBS.

MS:MS as a screening technology is gradually being supplanted by rapid DNA genotyping targeting the genetic variants associated with the conditions of interest. The ability to identify abnormal levels of metabolites, or DNA variants associated with the group of genetic conditions known as inborn errors of metabolism has outpaced the health care system's ability to

assemble the full evidence required to evaluate the benefits and risks of screening (Khoury, L. McCabe, and E. McCabe, 2003; Andermann, Blancquaert, Beauchamp, and Déry, 2008). The issues that have emerged there is no lack of effective treatments for some of the conditions that are screened for (e.g. Duchenne muscukar dystrophy (DMD)) (Cragun, DeBate, and Pal, 2014). Screening for these conditions may do more harm in terms of anxiety and cost of follow-ups for unproven treatment options and may not even be beneficial (Dhondt, 2010). A number of bodies have proposed modifictions to the original Wilson and Jungner criteria so that their applicability to genetic disorders is appreciated, as exemplified in Box 2.

Box 2: Crossroads 99 Modification of Wilson & Jungner Criteria (Becker et al., 2011):

- Knowledge of population and disease
    - Burden of target disease should be important
    - Target population or population at risk identifiable
    - Considerable level of risk or latent or preclinical phase
    - Natural course (from susceptibility to precursor, early disease, and advanced disease) should be adequately understood
- Feasibilities of screening procedures
    - Suitable test or examination
    - Entire screening procedure acceptable to the population
    - Screening should be a continuing process and should encompass all elements of screening procedure
- Interventions and follow-ups
    - Interventions that have physical, psychological, and social net benefit available
    - Facilities for adequate surveillance, prevention, treatment, education, counselling, and social support available
    - Consensus on accepted management for those with positive test results
- Sociatal and health system issues
    - Costs should be balanced in economic, psychological, social, and medical terms and with health-care expenditures as a whole
    - Appropriate screening services accessible to the entire population, without adverse consequences for non-participants
    - Appropriate confidentiality procedures and antidiscrimination provisions for participants and non-participants

1.3 Informed decision making and consent in NBS

As well as the intention of improving health outcomes by correctly identifying disease at a stage when treatment is effective, NBS carries the potential for harms or negative consequences for infants and families. These include the impact of initial false positive screening test results; identifying a significant health condition for which no effective treatment is available; and identifying a genetic or metabolic anomaly for which the natural history is unclear but for which the child may be indefinitely followed up (also referred to as "over-diagnosis") (Miller et al. 2015).

In some health jurisdictions, NBS for certain diseases and conditions are mandated by the state, i.e. parents cannot decline. Screening for PKU is mandatory in one Canadian province, since the policy decision-makers have concluded that the evidence of benefit of early intervention for PKU is so clear that parents should not be in a position to deny this. However, in all other Canadian provincial jurisdictions, the legal basis for NBS is by parental consent. There is no guidance to professionals or NBS programs on how this should be implemented.

In practice, Canadian NBS programs seem to practise under a *de facto* opt out approach (Potter et al., 2015), in which parents are assumed to consent if they raise no objections. However, research by the project team for this study (Etchegary et al., 2016; Nicholls et al., 2019), and others, indicates that some parents appear unaware that screening even occurred, and others feel that they were not consulted to the extent they would have preferred.

Without the need to actively seek consent, there is no obligation on health care providers to provide information unless requested, and lack of clarity about who has primary responsibility for doing so. Ulph, Dharni, Bennett, and Lavender (2019) suggest that most parents accept assumed consent if adequately informed, but that some may change their opinion once aware of

certain aspects of screening (e.g., who has access to their child's specimen after screening is complete). If not properly informed, parents may be unpleasantly surprised by an abnormal screening result for ther child, and may experience more anxiety than those who understood the possibility of false positives. Additionally, a parent might wish to decline screening if it meant that they would find out their child had an untreatable condition.

In addition to ethical considerations, there are positive arguments to support ensuring a consent process that effectively meets parents' expectations. The findings of Etchegary et al. (2016) suggested that parents whose infants had negative (normal) screening results tended to be more accepting of less explicit consent processes, whereas parents who had declined screening, or those who received their maternity care from midwives had higher expectations that NBS consent processes should be more explicit and informed.

The focus of this thesis was to understand parents' preferences for the consent procedure of NBS. Designing a consent process for NBS that effectively meets parents' expectations would need to consider variation in expectations and preferences between parents, the potential for a consent process to be perceived to be overly intrusive in the context of pregnancy and delivery, and the opportunity costs to the healthcare system of introducing significant change. Hence, this project was designed to address the research question "How can we build a consent procedure for NBS that meets parents' expectations?" Answering this question requires understanding what parents prefer in the NBS consent procedure. The objective of this study is to assess parents' past experience of NBS and attitude towards potential consent procedures of NBS; quantifying parents' preferences for key attributes of NBS consent procedures; and identify the subgroups of different preferences and expectations of how consent for NBS should be carry out. To achieve these objectives, a survey was designed to collect parents' past experiences of newborn

screening, attitude towards potential newborn screening consent procedure, and various opinions towards the government and healthcare system. A discrete choice experiment (DCE) was designed as a part of the survey to collect parents' preferences of newborn screening consent procedures.

CHAPTER 2 Methods

Preamble

This study was part of a multi-stage project lead by a research with members in University of Ottawa, The Ottawa Hospital Research Unit, and Memorial University of Newfoundland. The work presented in this study formed a part of the final stage of the project, focusing on the analysis of discrete choice experiment (DCE) component of a cross-sectional population-based survey.

The overall approach to the instrument design and data collection methods were developed by the full research team. The following activities formed the thesis research:

1. Designing the cross-sectional survey and DCE for data collection.
2. Development of the analysis strategy
3. Data cleaning
4. Analyses: primary descriptive, DCE, latent class analysis.

Section 1 describes the project activities carried out by the research team in advance of the thesis research. They are included here for completeness, and so that specific aspects of the design and methods may be taken into consideration in the evaluation of the findings and interpretation presented in this thesis.

2.1 Section 1: Cross sectional Survey Instrument

2.1.1 Approach to Development

The goals of the survey were to capture data on:

1. Demographic attributes of participants

2. the past personal experiences of consent processes associated with NBS for their youngest child
3. attitudes to authority and consent for NBS procedures, and personal preferences for how consent processes should be carried out in practice
4. individual preferences underpinning personal health-related decision-making in general
5. attitudes towards (including trust in) government and health care authorities
6. preferences for combinations of specified, distinct elements of a hypothetical NBS consent process

As well as allowing for descriptive analyses in their own right, the categories of data captured in domains 1-5 were specifically developed so that they could be incorporated into the analysis of data in domain 6, providing additional insights into characteristics associated with different preferences for how NBS consent should be handled. This is explained in further detail in section 2.

The survey study was designed primarily by the Ottawa research team, with contribution of this thesis research to the development and refinement of the final section (DCE), described in Section 2 below. The survey was administered in a cross-sectional design. Prior to this thesis research, the research team drew on the findings of earlier stages of the larger project (Etchegary et al., 2016; Nicholls et al., 2019) and conducted targeted literature searches to identify existing scales, instruments, and survey items that related to the survey goals. If no relevant instrument was found in a particular area, the research team developed and tested their own bespoke items.

The draft survey thus created was reviewed by a convenience sample of five participants for language clarity, completion feasibility, including the time required. Minor revisions only were made.

The final instrument (Appendix 5.1) consisted of items that captured data in six domains.

1. Participant demographics (developed by research team)
2. Recall of personal experience of NBS consent (developed by research team)

This was developed by the research team based on the .

3. Opinions about how newborn screening should be provided by provinces (developed by research team)
4. Individual decision-making preferences (developed by research team)

The research team selected two instruments: the Decision Making Control Instrument (DMCI) (Miller et al 2011) and the Health Information Orientation Scale (HIOS) (Dukenske et al 2009).

The DCMI measures the 'voluntariness' of respondents' decision-making (Miller et al 2011). The instrument comprises nine statements (see Appendix Table 5.1), each with a response scale from 1 (strongly disagree) to 6 (strongly agree). After reversing those for statements 3 and 9, the scores are added to give a total score in the range 1-36. Higher total scores indicate higher levels of overall individual 'authority' in decision-making on the part of respondents.

Three subscales can be derived from the DCMI, based on summing the scores for three items each:

- self-control (statements 3, 8, 9): indicates how well individuals can make the decision for themselves - higher scores indicate stronger ability to make the decision.

- absence of control (statements 1, 4, 6): indicates the level of control individuals feel during the decision-making process - lower scores indicate sense of individual lack of control during the decision-making process.
- others' control (statements 2, 5, 7): indicates extent to which decisions can be influenced by others - higher scores indicate more resistance to the influence of others.

The HIOS (Dukenske et al 2009) measures respondents' tendency to seek or avoid information in relation to making decisions. The instrument comprises eight statements (see Appendix Table 5.2), each with a response scale scored from 0 (not true at all) to 4 (very much true). Four statements (1-4) represent an 'information engagement' subscale, and four (5-8) represent an 'information apprehension' subscale. For each subscale, the scores are averaged, with a higher score indicate stronger willingness to seek, or avoid, information, respectively.

5. Trust in government and the healthcare system (developed by research team)

The research team selected the Culture World View Scale (Lachapelle et al 2014), which measures respondents' opinions of the government's role in individual's decision-making. The scale comprises eight statements (see Appendix Table 5.3), forming three subscales. Each statement has a response scale scored from 1 (strongly disagree) to 4 (strongly agree).

The three subscales are:

- Hierarchism (statements 1-3)
- Individualism (statements 4-6)
- Egalitarianism (statements 7, 8).

For each subscale, the scores are averaged. Higher scores indicate stronger alignment with the represented perspective.

6. pairwise choice tasks for discrete choice experiment. This domain is described in Section 2 below.

2.1.2 Participants and recruitment

Study Population and Sample

The approach and methods were developed by the research team.

The target population was all parents to whom NBS was offered in Canada since tandem mass spectrometry was introduced as a routine screening technology for metabolic and related disorders in newborn infants.

The study population comprised individuals identifying as parents of children who were born in Canada in the preceding five years, and who were registered as general access panel members with the public attitudes research company Dynata (https://www.dynata.com/). Dynata recruits participants across the country through partnerships with social media, blogs, online communities, and various websites. Upon joining the panel, participants must agree to Dynata's privacy policies, confidentiality terms and obligations, and the relationship of the contractual nature.

The study sample comprised Dynata panel members who fulfilled the following eligibility criteria:

Inclusion:

- aged at least 18 years old
- able to read and write in English or French
- have at least one child aged 5 years or younger who was born in Canada

A quota sampling method was used. Dynata identified the pool of panel members whose profiles indicated potential eligibility for the survey, and messaged them through email or text

message. The eligibility criteria were presented as four initial screening questions. Respondents who answered 'no' to any one of the four questions were excluded at that stage. Recruitment remained open until the target sample size was achieved, at which point the survey was closed. The sample size of this study was guided by the simulation from Dziak, Lanza, and Tan (2014). The article suggested that 600 samples would be needed if we expect to discover multiple classes within the sample. However, the simulation in Dziak, Lanza, and Tan (2014) may be underpowered occasionally. Hence, taking the more conservative approach, the research team aimed to achieve a final sample size of at least 700 samples. A total of 715 samples were included in this study.

2.1.3 Discrete Choice Experiment

The DCE component of the survey was designed to quantify how participants valued different attributes of a hypothetical NBS consent process. Although other domains of the survey also contain items addressing preferences and opinions about NBS consent processes, they do not allow an analysis of the relative value or importance of these attributes, nor can they shed light on how these attributes might be traded off against each other. In real life, a product or service comprises several different features, and rarely do all of these features come together in an optimal way for the consumer or client. For example, purchasing a new car may require the buyer to decide where to compromise between three attributes – fuel efficiency (good vs. better vs. best), interior space (adequate vs. spacious), and length of warranty coverage by the manufacturer (shorter or longer) – the individual has to choose the most ideal combination of attribute levels (in parenthesis) instead of the ideal level for each attribute individually.

The principle of a DCE exercise is that 'choice sets' are created by 'conjoining' the attributes of interest; the levels being set by the designer. Participants are presented with a

number of choice sets and required to select the one they prefer. This ‘choice task’ therefore forces the respondent to trade the attributes off against each other and, thus, reveals their relative value to the respondent. This method was used to examine the preferecnes of parents’ preferences for Down’s syndrome screening test by Carroll, Al-Janabi, Flynn, and Montgomery (2013) and quantify patients’ preference strength in multiple aspects of early assisted discharge by Goosen et al. (2014).

A DCE can provide useful insights to guide policy or practice; in this study, the goal was to quantify the relative value of different aspects of a consent process in order to inform the re-design of the process, if applicable.

To develop the DCE section, the entire research team worked together. Based on a review of the findings of earlier stages of the parent research study, a long list of potential attributes was developed. This list was refined through discussion, with the goal of retaining no more than 4-5 attributes, to ensure that the choice task was not unreasonably burdensome. The team also focused on selecting attributes, and their levels, characteristics that were potentially modifiable within a program context. Over several iterations, the number of attributes was reduced to four, along with the fewest levels per attribute. The final set of attributes and their accompanying levels is summarised in Table 2.1a.

Table 2.1a: Attributes and levels

Attribute	Levels
Best time to introduce information	Earlier in pregnancy (first or second trimester)
	Later in pregnancy (third trimester)
	At the time of the actual heel prick when the sample is collected
Healthcare professional's role	To provide a clear recommendation about screening to parents
	To work with parents to come to a mutually agreeable decision
	To provide sufficient information for parents to make their own decision
Reasonable assumption about consent	Consent can reasonably be assumed unless the parent says otherwise
	Consent can never reasonably be assumed and should always be discussed with the parent
Consent documentation	It is seldom necessary to record the parents' consent decision in writing
	It is not necessary to record the parents' consent decision in writing unless screening is declined
	It is always necessary to record in writing the parents' consent decision

For this survey, we created a set of pairwise DCE tasks, i.e., for each task, respondents were asked to choose between two alternatives. Table 2.1b provides an example.

Table 2.1b: DCE Choice Task Example

	Scenario A	**Scenario B**
Best time to introduce information	Earlier in pregnancy (first or second trimester) so there's plenty of time to think about it	Later in pregnancy so it's closer to the birth and more relevant to making the screening decision
Health professional role	To provide a clear recommendation about screening to parents	To provide sufficient information for parents to make their own decision.
Reasonable assumption about consent	Consent can reasonably be assumed unless the parent says otherwise.	Consent can reasonably be assumed unless the parent says otherwise.
Consent documentation	It is seldom necessary to record the parent consent decision in writing	It is always necessary to record in writing the parent consent decision
CHOOSE A or B	[ ]	[ ]

A 'full' balanced DCE for the selected attributes and levels would require a total of $3^3 2^1 = 54$ tasks (Kuhfeld, 2010) – three attributes have three levels and one attribute has two levels. In order to reduce respondent burden, this was reduced to a set of 12 DCE tasks per participant. Making use of the technical ability of an online survey tool, a total of 300 different versions of task sets were created, and 12 were presented randomly to each participant.

The overall preference pattern of the attributes would be examined using the conditional logit model and the Latent Class (LC) Regression model would be used to assess how preferences may vary among different subgroups.

2.2 Section 2: Data Analysis

2.2.1 Analysis of Cross sectional Survey

The purpose of descriptive analysis was to determine who are the participants of the survey and accesses participants' past experience of and attitude towards newborn screening. The responses in domains 1-5 were summarized using frequency analysis.

2.2.2 Analysis of DCE data

Conditional Logit Regression

The conditional logit model is most commonly used to analyze DCE (Kuhfeld, 2010). The conditional logit model was appropriate for the analysis because the outcome variable of DCE is usually in a one to one or one to many matched format. In the current situation, out of each pairwise task, the scenario chosen would be considered a case (outcome of 1) and matched against a control (outcome of 0), which would be the scenario not chosen. The conditional logit model would be used to examine the overall preference pattern of attributes in the sample. The absolute values of β estimates of the conditional logit model, also known as the preference weight, alone has no meaningful interpretation since preference weights only measure the relative preference level (Hauber et al., 2016). However, the absolute value of difference between preference weights measures the importance of the attribute. This difference represents the amount of utility change when moving from one level to another within the attribute (Hauber et al., 2016). The attribute with the highest preference weight difference was deemed the most important attribute.

One drawback of the conditional logit model is that it assumes that the average preference weights are the same across the entire sample (Hauber et al., 2016). The research

team however, were interested in exploring whether subgroups of respondents had different average preferences, which required a different statistical approach.

Latent Class (LC) Regression

A LC regression model assumes the existence of one or more latent (or 'hidden') classes within the sample, with different preference patterns (Hauber et al., 2016). This approach requires the number of classes to be determined before running the regression analysis. Hauber et al. (2016) suggest that Akaike Information Criterion (AIC) or Bayesian Information Criterion (BIC) can inform the choice of the number classes that should be included in the model. However, using AIC solely may result in overfitting with too many classes while using BIC solely may result in underfitting (too few classes) (Hauber et al., 2016). Noting these caveats, the research team judged that the number of class would be decided after inspecting the initial results, expecting that a 3- or 4-class model would be the likely target given prior understanding of the study context. Thus, we planned a regression model estimated for each class using the EM or Newton Raphson Algorithm (Vermunt & Magidson, 2002). The Stata command *lclogit* described by Pacifico and Yoo (2013) was utilized to run the LC Regression model for this study.

Class Membership Association

After the LC regression, we would like to figure out the characteristics of each class. We have chosen several variables collected from the survey and examine their association with the class membership. We have convert the category variables selected into binary variables for simplicity. Table 2.2a and b shows the variables that we believe are the best to describe the characteristics of each class. Since we were expecting a 3- or 4-class model from the LC regression, the multinomial logit model would be appropriate for modeling the association of

class memberships. The multinomial logit model is similar to the regular logit model (logistic regression). However, the outcome variable of multinomial logit model has more than two categories while the outcome variable of the regular logit model is binary.

Table 2.2a: Selected categorical variables

Variables	**Categories**
Demographics	
Age	Under 35 years old
	35 years old or over
Gender	Male
	Female
Highest education	Bachelor's degree or higher
	Less than Bachelor's degree
Number of children at home	Two or more
	One
Healthcare professional status	Yes
	No
Past Experience and opinions of NBS	
Sufficiency of information provided	Just enough or too much information provided
	No information provided or too little
Amount of time that should be given for the parent to decide	More than a week
	A week or less
Parents should have to actively choose whether to screen their children or not	Slightly agree or agree
	Slightly disagree or disagree
Parents should not be allowed to say no to screening	Slightly agree or agree
	Slightly disagree or disagree
Trust in healthcare system	
The hospital only provides screening tests that are important and safe	Agree or strongly agree
	Neutral, disagree, or strongly disagree

Table 2.2b: Selected continuous variables

Variables	Min	Max
Decision Making Control Instrument		
Total DMCI mean score	9	54
Health Information Orientation Scale		
Information Engagement Information Apprehension	0 0	4 4
Socio-culture Perspective		
Hierarchism Individualism Egalitarianism	1 1 1	4 4 4

2.3 Section 3: Ethical Aspects

This study was approved by the Ottawa REB and the Memorial University Health Research Ethics Board (HREB). The data for this thesis were provided under a data sharing agreement between the University of Ottawa and Memorial University of Newfoundland (Appendix 5.3). The data were stored in a secured cloud drive, with access restricted to the Memorial investigators and the candidate.

The survey data were de-identified by Dynata before being supplied to the University of Ottawa investigators. Within the dataset, each participant was identified by a unique ID which bore no relation to their actual identity. No personal information such as residential address, postal code, birthdates, or even province of residence were supplied to the investigators in the survey.

CHAPTER 3 Results

3.1 Part 1: Descriptive Analysis

3.1.1 Participant Demographics

Table 3.1 (following page) presents the characteristics of the 715 survey respondents. To provide a comparison with the Canadian population within the same age group, data from the 2016 census (Statistics Canada, 2017) were reviewed. The majority of the sample identified as white (69.5% vs. 57.1% with European origin in the 2016 census), relatively young (60.4% aged 30 to 39 v 18.1% in the 2016 census), and majority female (58.9% v 51.5% in the 2016 census). Of those who disclosed it, 66 (9.4% v 6.2% in the 2016 census) identified as Indigenous. About half had at least an undergraduate degree (50.5% v 23.3% in the 2016 census), and the proportion with an after-tax household income above the median ($62,900, according to Statistics Canada (2021)) was (58% vs 50% in the 2019 Canadian Income Survey). A total 97 out of 712 participants (13.6%) identified themselves as healthcare professionals.

Around half of the participants reported they had only one child (49.9%), and the place of birth for their youngest child was a hospital (90.7%).

Table 3.1: Characteristics of survey respondents

Demographics	Categories	N	Responses
Age N (%)	≤ 24 25 to 29 30 to 34 35 to 39 ≥ 40	713	36 (5.1) 111 (15.6) 237 (33.2) 194 (27.2) 135 (18.9)
Gender N (%)	Male Female Other	713	291 (40.8) 420 (58.9) 2 (0.3)
Indigenous status N (%)	Indigenous Not Indigenous Prefer not to answer	711	66 (9.3) 633 (89.0) 12 (1.7)
Ethnicity N (%)	White South Asian Chinese Black Arab Other	712	495 (69.5) 51 (7.2) 39 (5.5) 30 (4.2) 17 (2.4) 80 (11.2)
Marital status N (%)	Married or living as married Widowed Divorced Single-never married	706	631 (89.4) 7 (1.0) 13 (1.8) 55 (7.8)
Annual income ($) N (%)	≤ 25,900 25,901 to 46,100 46,101 to 70,800 70,801 to 111,500 > 111,500	713	42 (5.9) 79 (11.1) 179 (25.1) 240 (33.7) 173 (24.3)
Highest education level N (%)	No post-secondary Trade certificate or diploma Community college University certificate below Bachelor's level Bachelor's Degree Post-graduate degree	713	79 (11.1) 71 (10.0) 104 (14.6) 99 (13.9) 299 (32.1) 131 (18.4)
Identifies as a health professional N (%)	Yes No	712	97 (13.6) 615 (86.4)
Geographical residence N (%)	Rural Small city/town Medium or large city	711	95 (13.4) 131 (18.4) 485 (68.2)
Language spoken at home N (%)	English French Other	709	658 (92.8) 26 (3.7) 25 (3.5)
Number of children in household N (%)	One Two or more	711	355 (49.9) 356 (50.1)
Youngest child's place of birth N (%)	Hospital Birthing centre Home	712	646 (90.7) 49 (6.9) 17 (2.4)

3.1.2 Recall of how NBS was presented

Tables 3.2a and b summarise respondents' recall of information provision and consent for NBS for their youngest infant. The majority of participants reported that the most frequent source of information about NBS was a healthcare provider (70.7%), mostly verbally (55.4%), sometimes accompanied by written material. Of the 540 respondents who could remember receiving information, just over half reported being informed about this during pregnancy; a small proportion reported that they were informed for the first time when the sample was about to be taken (12.8%) or after it had been collected (3.9%).

Of the 544 who reported receiving information, two thirds indicated that the amount of information was about right (62.7%), and most of the others felt it was too little (33.1%). Of 708 who responded, 402 (56.8%) of them recording reading the NBS information provided. Although information was provided, over half of participants (> 60%) did not understand the provided information except for that pertaining to the mode of sample extraction (50.9%).

Around two thirds of repondents (61.7%) reported that they recalled being asked to consent to NBS for their infant (Table 3.2b).

Table 3.2a: Recall of NBS information provision for youngest child

Aspect	Categories	N	Responses
How information was provided by healthcare providers N (%)	Not provided or cannot remember Provided verbally only Provided in written format only Provided both verbally and in written format	713	209 (29.3) 279 (39.1) 109 (15.3) 116 (16.3)
Earliest time when information was provided N (%)	Early in pregnancy Late in pregnancy When the baby was born When sample was taken After sample was taken Cannot remember or no information received	714	149 (20.8) 132 (18.5) 169 (23.7) 69 (9.7) 21 (2.9) 174 (24.4)
Sufficiency of information provided N (%)	No information received Too little Just right Too much	713	169 (23.7) 180 (25.3) 341 (47.8) 23 (3.2)
Understanding of information content N (%)			
	Name of conditions screened for	715	236 (33.0)
	How to prepare baby for test		235 (32.9)
	How the sample would be taken		364 (50.9)
	Possible pain or side effects when the sample is taken		254 (35.5)
	Time to receive results		220 (30.8)
	How the results would be communicated		201 (28.1)
	Possibility of false negative results		114 (15.9)
	Possibility of false positive results		87 (12.2)
	Health effects of conditions being screened for		130 (18.2)
	Rarity of conditions being screened for		137 (19.2)
	Treatment options for conditions being screened for		96 (13.4)

Table 3.2b: Recall of consent timing

Recall of when consent was sought	N	Responses
Early in pregnancy Late in pregnancy When the baby was born When the sample was taken After the sample was taken Does not recall when was consent asked Never asked	713	90 (12.6) 101 (14.2) 176 (24.7) 84 (11.8) 21 (3.0) 102 (14.3) 139 (19.5)

3.1.3 Views on NBS consent practicalities

Table 3.3a summarizes responses to questions about when and how information should be provided to parents, when the decision to screen or not should be made, and the need for documentation.

In terms of NBS information provision, participants indicated multiple methods of providing information as useful, of which discussion with a healthcare professional was selected as the most useful by 45.4%. For timing, after excluding 39 who indicated that consent should be compulsory, 49 (7.3%) suggested that the screening decision could be made at the time when sampling would be done, the remainder roughly even split between early pregnancy, late pregnancy, and around the time of birth. For the time required by parents to make a decision, and excluding the 61 who indicated that screening should be compulsory, 260 (39.9%) felt that at least a week should be allowed. Of the 668 who expressed a view, 538 (80.5%) indicated that screening could proceed only after parental agreement was documented by either the provider or a parent.

As indicated in Table 3.3b, participants appeared to very definitively indicate views that healthcare providers have a duty to provide information, parents have a duty to review it, and that their understanding of it should be checked before screening is actually carried out.

Table 3.3a: Views on NBS information provision for parents

Aspect	Categories	N	Responses
Earliest time when NBS information should be provided to parents N (%)	Early in pregnancy Late in pregnancy When the baby is born When sample about to be taken After the sample is taken	715	323 (45.2) 272 (38.1) 90 (12.6) 22 (3.1) 7 (1.0)
Ways of providing information that would be useful N (%)[1]	Through discussion Leaflet Internet Mobile/tablet app Group session	715	525 (73.4) 435 (60.8) 321 (44.9) 185 (25.9) 94 (13.2)
The single most useful way of providing information N (%)	Through discussion Leaflet Internet Mobile/tablet app Group session	710	322 (45.4) 176 (24.9) 134 (18.9) 52 (7.3) 26 (3.6)
When the screening decision should be made N (%)	Early in pregnancy Late in pregnancy When the baby is born When the sample is about to be taken Screening should be compulsory	714	183 (25.6) 238 (33.3) 205 (28.7) 49 (6.9) 39 (5.5)
Amount of time that should be given for the parent to decide N (%)	Screening not subject to parental decision Long enough to read the leaflet Time to discuss in the moment A week or two More than two weeks	713	61 (8.6) 186 (26.1) 206 (28.9) 204 (28.6) 56 (7.9)
Condition for allowing screening to proceed N (%)	Verbal agreement alone Verbal agreement, recorded by provider Written agreement by parent If no parental objection, may be assumed Does not really matter	711	92 (12.9) 259 (36.4) 279 (39.2) 38 (5.3) 43 (6.1)

[1] Multiple responses

Table 3.3b: Views on ensuring that information is provided and understood

Responsibility	N	Agree with stagement
Healthcare professionals have a responsibility for providing information about NBS	712	683 (95.9)
Parents have a responsibility to review information materials provided to them about NBS	709	658 (92.8)
A parent's understanding of key pieces of information should be confirmed before screening proceeds	710	658 (92.7)

3.1.4 Views on voluntariness of screening

Going down the rows in Table 3.4, the items move loosely from a consent approach in which parents are expected to make a decision about NBS themselves, through to a mandatory screening approach. Overall, over 75% of respondents tended to agree with each statement, with the exception of the final one - that parents should not be allowed to decline NBS, for which responses were evenly split between agreement and disagreement.

Table 3.4: Views on the extent to which screening should be voluntary or compulsory

Statement	N (%) 1=agree, 4=disagree				
	N	1	2	3	4
Parents should have to actively choose whether to screen their children or not	713	395 (55.4)	241 (33.8)	65 (9.1)	12 (1.7)
Parents should be strongly advised to have screening for their baby	713	289 (40.5)	297 (41.7)	88 (12.3)	39 (5.5)
Screening should happen unless a parent objects	712	265 (37.2)	270 (37.9)	136 (19.1)	41 (5.8)
Parents should be made to sign a disclaimer if they choose not to have their baby screened	712	354 (49.7)	260 (36.5)	69 (9.7)	29 (4.1)
Parents should not be allowed to say no to screening	712	131 (18.4)	221 (31.0)	217 (30.5)	143 (20.1)

3.1.5 Preferences and feelings about personal NBS decision-making

When asked a direct question about their preferred role in NBS decision making, Majority of participants (89.1%) want to make the decision themselves or have a shared responsibility with their healthcare providers. Most of participants prefer to make the decision after serious considering provider's opinion (45.8%). (see Table 3.5).

Table 3.5: Preference for NBS decision making

Preference	N	Responses (%)
Make the decision about screening entirely by myself	714	152 (21.3)
Make the decision after seriously considering provider's opinion		327 (45.8)
Share decision-making responsibility with provider		157 (22.0)
Provider makes decision after seriously considering my opinion		47 (6.6)
Leave the decision entirely to provider		31 (4.3)

Tables 3.6a and b provide further insight into participants' feelings about the NBS decision for their youngest child, as assessed using the Decision Making Control Instrument (DMCI) (Miller et al., 2011). Higher scores indicate a relatively high level of 'voluntariness' in making the decision, as assessed by participants. The average total score of the overall sample was 35.7, above the mid-point of the scale (31.5), (Table 3.6b). Inspecting the individual items (Table 3.6a), 30-50% of participants agreed to some extent that they had not felt the decision to have their infant screened was completely voluntary.

Table 3.6a: Feelings about their experience of NBS decision-making (DMCI (Miller et al., 2011)), individual items

Statement		N	N (%) 1=strongly disagree, 6=strongly agree					
			1	2	3	4	5	6
1.	I was powerless in the face of this decision	710	115 (16.2)	149 (21.0)	140 (19.7)	145 (20.4)	101 (14.2)	60 (8.5)
2.	Someone took the decision away from me	709	156 (22.0)	163 (23.0)	148 (20.9)	112 (15.8)	83 (11.7)	47 (6.6)
3.	I made the decision	710	74 (10.4)	60 (8.5)	91 (12.8)	155 (21.8)	196 (27.6)	134 (18.9)
4.	I was passive in the face of this decision	707	83 (11.7)	116 (16.4)	152 (21.5)	197 (27.9)	103 (14.6)	56 (7.9)
5.	The decision was inappropriately influenced by others	709	145 (20.5)	171 (24.1)	149 (21.0)	136 (19.2)	64 (9.0)	44 (6.2)
6.	I was not in control of this decision	711	134 (18.9)	153 (21.5)	143 (20.1)	136 (19.1)	87 (12.2)	58 (8.2)
7.	Others made the decision against my wishes	707	191 (27.0)	159 (22.5)	134 (19.0)	113 (16.0)	65 (9.2)	45 (6.4)
8.	I was not the one to choose	708	143 (20.2)	144 (20.3)	128 (18.2)	138 (19.5)	97 (13.7)	57 (8.1)
9.	The decision was up to me	707	61 (8.6)	61 (8.6)	92 (13.0)	186 (26.3)	183 (25.9)	124 (17.5)

The three sub-scales each have a maximun score of 18 and minimun score of 3, with higher scores indicating greater voluntariness. For all three subscales, ('self-control' subscale, measuring respondents' feelings that they were able to make the final decision themselves; 'absence of control' subscale, measuring respondents' feelings that they were in control of the decision; and the 'others' control' subscale, measuring the extent to which respondents felt they were able to resist the influence of others in the decision), the middle point between the maximum score of 18 and minimum score of 3 was 10.5.

Table 3.6b: Feelings about their experience of NBS decision-making process (DMCI (Miller et al., 2011)), scale and sub-scale scores

Scale	Items	N	Mean	SD[1]	Min	Max
Total Score		694	35.7	9.87	9	54
Self-control	3, 8, 9	705	12	3.70	3	18
Absence of Control	1, 4, 6	707	11.3	3.71	3	18
Others' Control	2, 5, 7	702	12.4	4.04	3	18

[1] Standard Deviation

3.1.6 Preferences about personal medical decision making in general

Tables 3.7a and b present data on participants' general approach to making decisions about their healthcare, assessed using the Health Information Orientation Scale (DuBenske et al 2009). This measures the tendency of a respondent to seek or avoid information when making decisions.

Taken overall, the distribution of responses for the individual items (Table 3.7a) indicates, as a group, a tendency towards gathering, reviewing, and continuing to seek information; not making decisions quickly; being able to make sense of information from multiple sources, of coping with a large amount of information, and of not being afraid of learning something unexpected; and feeling that it's their job (not the provider's) to deal with information.

The maximum sub-scale scores are 4, with a higher score indicating higher engagement or apprehension, respectively. The scores for both the information engagement and apprehension sub-scales (See Table 3.7b) are consistent with a tendency to interact actively with health information. The wide standard deviations around the apprehension sub-scale suggests that a proportion of respondents feel less competent in working with health information (DuBenske et al 2009).

Table 3.7a: Information to support decision-making, in general (Health Information Information Orientation Scale (Dukenske et al., 2009)), individual items

Statement		N	N (%) 0=not true at all, 4=very much true				
			0	**1**	**2**	**3**	**4**
1.	I like to gather as much information as I can before making a decision	712	13 (1.8)	49 (6.9)	130 (18.3)	313 (44.0)	207 (29.1)
2.	I review information multiple times before making a decision	709	9 (1.3)	79 (11.1)	199 (28.1)	268 (37.8)	154 (21.7)
3.	After I made a decision, I continue to look for related information	711	35 (4.9)	116 (16.3)	223 (31.4)	221 (31.1)	116 (16.3)
4.	I like to make decisions quickly	710	137 (19.3)	165 (23.2)	216 (30.4)	137 (19.3)	55 (7.8)
5.	I have difficulty make sense of information from multiple sources	710	200 (28.2)	157 (22.1)	186 (26.2)	120 (16.9)	47 (6.6)
6.	I fear I might find out something I don't want to know	711	146 (20.5)	168 (23.6)	197 (27.7)	144 (20.3)	56 (7.9)
7.	I feel overwhelmed by the amount of information available	711	136 (19.1)	189 (26.6)	185 (26.0)	152 (21.4)	49 (6.9)
8.	I think it's the doctor's job to deal with information, not mine	710	261 (36.8)	154 (21.7)	157 (22.1)	89 (12.5)	49 (6.9)

Table 3.7b: Information to support decision-making, in general (Health Information Orientation Scale (DuBenske et al., 2009)), sub-scale scores

Scale	Items	N	Mean	SD[1]	Min	Max
Information Engagement	1-4	712	2.42	0.17	0	4
Information Apprehension	5-8	712	1.56	1.01	0	4

[1] Standard Deviation

When asked directly, 169 of 709 (23.8%) participants indicated that religious or spiritual beliefs influence their medical decision-making.

3.1.7 Views on the provision of healthcare

Table 3.8 summarises responses to items designed to elicit views on trust and confidence in healthcare as it relates to NBS. Overall, the responses indicated at least fair confidence in the safety and benefits of tests that are offered or paid for through government funding, the quality of the healthcare system, and the motivations of medical researchers. The most mixed responses related to the need for double-checking on interventions carried out on their children.

Table 3.8: Views on trustworthiness of healthcare

Statement	N	N (%) 1=strongly disagree, 5=strongly agree				
		1	2	3	4	5
The government will ensure a high quality healthcare system	711	51 (7.2)	80 (11.3)	198 (27.8)	268 (37.7)	114 (16.0)
I feel like I have to double check everything the hospital does to my baby	710	44 (6.2)	153 (21.6)	214 (30.1)	208 (29.3)	91 (12.8)
The hospital only provides screening tests that are important and safe	711	16 (2.3)	43 (6.1)	208 (29.3)	319 (44.9)	125 (17.4)
Most medical researchers want to work on things that will make life better for the average person	709	13 (1.8)	33 (4.7)	179 (25.3)	344 (48.5)	140 (19.8)
If the government has funded a health test or procedure it's probably a worthwhile test to have	711	21 (3.0)	37 (5.2)	224 (31.5)	283 (39.8)	146 (20.5)
The government wouldn't fund a test or procedure if they were not sure of its benefits	711	17 (2.4)	70 (9.8)	224 (31.5)	284 (39.9)	116 (16.3)

3.1.8 Socio-cultural perspectives

Tables 3.9a and b present data on how respondents see the relationship between individuals, society and authority, as assessed using the adapted 'cultural worldview' measures (LaChapelle et al 2014). The responses to the individual items are summarized in Table 3.9a.

Table 3.9a: Cultural worldview (Lachapelle et al., 2014), individual items

Statement		N	**N (%)** 1=strongly disagree, 4=strongly agree			
			1	2	3	4
1.	Government should put limits on the choices individuals can make so they don't get in the way of what's good for society	710	95 (13.4)	187 (26.3)	320 (45.1)	108 (15.2)
2.	Government should do more to advance society's goals, even if that means limiting the freedom and choices of individuals	712	120 (16.9)	198 (27.8)	293 (41.2)	101 (14.2)
3.	Sometimes government needs to make laws that keep people from hurting themselves	710	20 (2.3)	107 (15.1)	378 (53.2)	205 (28.9)
4.	It's not the government's business to try to protect people from themselves	710	107 (15.1)	271 (38.2)	239 (33.7)	93 (13.1)
5.	Government should stop telling people how to live their lives	708	59 (8.3)	244 (34.5)	302 (42.7)	103 (14.5)
6.	Government interferes far too much in our everyday lives	710	78 (11.0)	289 (40.7)	249 (35.1)	94 (13.2)
7.	We need to dramatically reduce inequalities between the rich and the poor, as well as between men and women	711	29 (4.0)	118 (16.6)	302 (42.5)	262 (36.9)
8.	Our society would be better off if the distribution of wealth was more equal	711	34 (4.8)	128 (18.0)	309 (43.5)	240 (33.7)

The maximum score for each scale was 4. Overall responses indicated a sample whose worldview mapped most closely to valuring fairness and equity (egalitarianism scale), followed social order (hierarchism scale), and then freedom and autonomy (individualism scale). While the mean score for each scale indicates agreement with the proposed underlying worldview, inspection of the responses to individual items suggests heterogeneity in the disaggregated scores for the individualism scale.

Table 3.9b: Cultural worldview (Lachapelle et al., 2014), scale scores

Scale	Items	N	Mean	SD[1]	Min	Max
Hierarchism	1-3	712	2.74	0.69	1	4
Individualism	4-6	712	2.53	0.70	1	4
Egalitarianism	7, 8	711	3.09	0.75	1	4

[1] Standard Deviation

3.2 Part 2: Analysis of the Discrete Choice Experiment

3.2.1 Relative importance of each attribute

The results of the conditional logit regression analysis for the total group of respondents are summarized in Table 3.10. Based on a significance level of $\alpha = 0.05$, these suggest that three attributes have effects on the preferences of the overall sample: the best time to provide information about NBS, the reasonable assumption of consent, and the required consent documentation. Our data suggest that best time to provide information were the most valued attribute by the overall sample since it has the largest preference weight difference (0.51 (= 0.16 – [-0.35])) among all attributes. Participants prefer to receive information late in pregnancy, always ask for consent, and always document consent decisions. Participants also like to avoid only getting information after their babies are born and not recording consent decisions. On Average, participants were 1.18 times more likely to choose the option with information provided in late pregnancy (OR: 1.18); 1.13 times more like to choose the option with always ask for consent (OR: 1.13); and 1.28 times more likely to choose the option with the consent decision being documented (OR: 1.28). They were 0.7 times less likely to choose the option with information provided after the birth of their children (OR: 0.70) and 0.85 less likely to choose the option with the consent decision not being documented (OR: 0.85).

Table 3.10: Overall discrete choice model

Attribute	Level	Parameter Estimate	Odds Ratio	P-value
Best Time to Provide Information	Early in pregnancy	Ref	Ref	-
	Late in pregnancy	0.16	1.18	< 0.01
	After birth	-0.35	0.70	< 0.01
Healthcare professional's role	To provide a clear recommendation	Ref	Ref	-
	To work with parents to come to a mutually agreeable decision	-0.03	0.97	0.32
	To provide sufficient amount of information	-0.03	0.97	0.46
Reasonable assumption about consent	Can be reasonably assumed unless parents say otherwise	Ref	Ref	-
	Cannot be reasonably assumed	0.13	1.13	< 0.01
Consent Documentation	Seldom necessary to record consent decision in writing	Ref	Ref	-
	Not necessary to record consent decision	-0.16	0.85	< 0.01
	Always necessary to record consent decision	0.25	1.28	< 0.01

Table 3.10a: Goodness of fit statistics

Criterion	Model without covariates	With covariates
-2 log likelihood	11894.406	11493.391
AIC	11894.406	11507.391
BIC	11894.406	11556.792

3.2.2 Classes with differing preference patterns

Within the overall sample, latent class regression analysis identified three classes with differing preference structures. Their characteristics are compared in Tables 3.11a-c. Originally, the 4-class model produced the best AIC and BIC values. However, due to the extreme similarity between two of the classes in the 4-class model, we decided to use the 3-class model instead. The expected class memberships of each class are: 20.2% for class 1, 20.8% for class 2, and 59% for class 3.

For Class 1 (Expected 21.1% of sample), the same three attributes as observed for the total sample appeared to have statistically significant effects on the overall preference (Table 3.11a). Inspection of the odds ratios suggest the largest effects from the two consent-related attributes (reasonable assumption and documentation). Consent documentation was valued the most by participants in Class 1 with preference weight difference of 2.45 (= 1.59 – [-0.86]). In summary, this group appeared to somewhat prefer NBS information *not* be provided after the baby is born, to feel (more strongly than the total sample) that consent should not be reasonably assumed, and to feel very strongly that the consent decision should always be recorded. The participants in this group were 0.7 times less likely to choose the option with the information provided after birth (OR: 0.7); 2.4 times more likely to choose the option with consent not being assumed (OR: 2.40); and 4.52 times more like to choose the option with the consent decision always being documented (OR: 4.52). We labelled this class as the "consent priority" class (abbreviated to "consent" class in tables).

Table 3.11a: **Discrete choice model**, **class 1** ("consent", expected membership, 21.1%)

Attribute	Level	Parameter Estimate	Odds Ratio	P-value
Best Time to Provide Information	Early in pregnancy	Ref	Ref	-
	Late in pregnancy	0.30	1.35	0.06
	After birth	-0.36	0.70	0.02
Healthcare professional's role	To provide a clear recommendation	Ref	Ref	-
	To work with parents to come to a mutually agreeable decision	0.08	1.08	0.51
	To provide sufficient amount of information	0.10	1.10	0.44
Reasonable assumption about consent	Can be reasonably assumed unless parents say otherwise	Ref	Ref	-
	Cannot be reasonably assumed	0.88	2.40	< 0.01
Consent Documentation	Seldom necessary to record consent decision in writing	Ref	Ref	-
	Not necessary to record consent decision	-0.94	0.39	< 0.01
	Always necessary to record consent decision	1.51	4.52	< 0.01

For Class 2 (Expected 20.5% of the sample), one attribute appeared to largely drive the overall preference, the timing of information provision, with consent documentation of borderline statistical significance. Like the overall group, this class appeared to prefer receiving NBS information later in pregnancy, and it appeared to have a stronger preference not to receive the information after the infant's birth (see Table 3.11b). The participants in this group were 1.53 times more likely to choose the option with information being provided in late pregnancy (OR: 1.53) and 0.07 times less likely to choose the option with information being provided after birth (OR: 0.07). The results also suggested that this class preferred that consent decisions should always be documented. They were 1.37 times more likely to choose the option with the consent decision always being documented (OR: 1.37). We labelled this the "information priority" class (abbreviated to "information" in tables).

Table 3.11b: **Discrete choice model, class 2** ("Information", expected membership, 20.5%)

Attribute	Level	Parameter Estimate	Odds Ratio	P-value
Best Time to Provide Information	Early in pregnancy	Ref	Ref	-
	Late in pregnancy	0.43	1.53	< 0.01
	After birth	-2.65	0.07	< 0.01
Healthcare professional's role	To provide a clear recommendation	Ref	Ref	-
	To work with parents to come to a mutually agreeable decision	0.14	1.16	0.28
	To provide sufficient amount of information	0.12	1.12	0.37
Reasonable assumption about consent	Can be reasonably assumed unless parents say otherwise	Ref	Ref	-
	Cannot be reasonably assumed	-0.004	1.00	0.97
Consent Documentation	Seldom necessary to record consent decision in writing	Ref	Ref	-
	Not necessary to record consent decision	-0.16	0.84	0.20
	Always necessary to record consent decision	0.32	1.37	0.03

For Class 3 (Expected 55.4%% of the sample), none of the attributes examined were associated with the overall preference (Table 3.11c). We labelled this the "flexible" class.

Table 3.11c: **Discrete choice model, class 3** ("Flexible", expected membership, 58.44%)

Attribute	Level	Parameter Estimate	Odds Ratio	P-value
Best Time to Provide Information	Early in pregnancy	Ref	Ref	-
	Late in pregnancy	0.09	1.10	0.09
	After birth	0.10	1.10	0.12
Healthcare professional's role	To provide a clear recommendation	Ref	Ref	-
	To work with parents to come to a mutually agreeable decision	-0.08	0.92	0.10
	To provide sufficient amount of information	-0.07	0.93	0.14
Reasonable assumption about consent	Can be reasonably assumed unless parents say otherwise	Ref	Ref	-
	Cannot be reasonably assumed	0.01	1.01	0.85
Consent Documentation	Seldom necessary to record consent decision in writing	Ref	Ref	-
	Not necessary to record consent decision	-0.03	0.97	0.61
	Always necessary to record consent decision	-0.06	0.94	0.28

3.2.3 Class Characteristics

Table 3.12 summarizes the association of the selected characteristics with class membership, using the 'flexible' class as the reference. Broadly speaking, the "consent" group was statistically significantly less likely to identify as a healthcare professional, and more likely to feel that parents should have at least a week to make a decision about NBS. This group was likely to have a higher score on the DCMI scale (indicating a greater sense of personal 'authority' in the last decision they made about NBS), and (borderline) a higher score on the cultural worldview individualism scale, indicating a perspective that the government should refrain from interfering in people's lives (Lachapelle et al 2014).

The "information" group was statistically significantly less likely to feel that they had had enough information when making a decision about NBS for their last child, and more likely to feel that parents should be allowed at least a week to make the decision. They were less likely to be apprehensive about seeking information, and more likely to judge that screening tests being offered by a hospital would be safe and important. Gender was of borderline significance, with males less likely to be in this class.

Table 3.12: Association of selected characteristics with class membership

Aspect		Information		Consent	
		OR[1]	p > |Z|	OR	p > |Z|
Participant demographics					
Under 35 years old		0.77	0.26	1.30	0.25
Male		0.63	0.05	0.88	0.54
Bachelor's degree or higher		0.79	0.31	1.17	0.48
More than one child at home		1.10	0.67	0.80	0.31
Healthcare professional		0.57	0.14	0.48	0.04
Past Experience and opinions of NBS					
Felt enough information was provided for last NBS decision		0.57	0.02	0.70	0.11
Felt that more than a week should be given for parent to decide on NBS		3.42	< 0.01	2.16	< 0.01
Parents should have to actively choose to screen or not		1.24	0.61	0.98	0.94
Parents should not be allowed to say not to screening		0.73	0.18	0.72	0.13
Decision-making and information gathering					
DCMI[2] total mean score		1.01	0.41	1.04	<0.01
HIOS[3] information engagement scale mean score		1.23	0.22	1.11	0.51
HIOS information apprehension scale mean score		0.65	< 0.01	0.87	0.28
Trust in healthcare system and socio-culture perspective					
Hospital only provides safe, important screening tests (agree)		2.07	< 0.01	0.90	0.64
Cultural worldview scale mean scores	*hierarchism*	0.74	0.09	0.75	0.08
	egalitarianism	0.74	0.08	0.78	0.13
	individualism	1.25	0.14	1.35	0.05

1. OR: odds ratio
2. DCMI: Decision Making Control Instrument (Miller et al., 2011)
3. HIOS: Health Information Orientation Scale (Dukenske et al., 2009)

Chapter 4 Discussion

This survey was conducted by a research team as the final component of a research program exploring how different parties experience and evaluate the consent process for NBS in Canada. Findings from the previous components pointed to a difference in expectations between parents, health professionals, and policy decision-makers, with how consent is 'supposed' to work being quite different from how it is experienced in practice (Etchegary et al., 2016; Nicholls et al., 2019; Ulph, Dharni, Bennett, and Lavender, 2019). Overall, an important intended outcome of the research program is recommendations about how consent procedures for NBS could be improved to meet all parents' expectations better while still being practical to implement. The study had the following general goals: assessing parents' own experiences of NBS decision-making and consent for their child and their attitudes towards how it was done, their more general perspectives on decision-making, healthcare, and views on the role of authority; quantifying the values of key attributes of an NBS consent process; and gaining insight into characteristics associated with different NBS consent process preference patterns if any. This study is built on the assumption that measuring the 'average' preferences in a group might miss different patterns held by sub-groups and that these differences might be important for developing recommendations but would be difficult to predict in advance. This assumption led to the inclusion of a DCE based on a latent class analysis approach, identifying different preference patterns without pre-specifying more than the number of classes being sought. The survey was also designed to capture data about participants covering several areas that might underlie different preferences (although not formally testing hypotheses about them): demographic characteristics, personal experiences of and attitudes towards NBS, individual decision-making styles, sense of trust in healthcare, and more general view of society and authority.

Although the survey was administered using an online panel, so did not use a sampling frame directly linked with NBS programs, the eligibility criteria should have ensured that participants reflected the population of parents who ought to have been informed about NBS for their youngest child. The project activities that fell specifically within the scope of the thesis research began with collaborating in developing the DCE section of the questionnaire, developing the complete analysis plan, and conducting and interpreting all of the analyses.

From the descriptive data, it appeared that a fifth of participants did not recall having been asked to consent to their infant being screened and a proportion only lightly lower than this were asked only at the time the sample was taken or even afterwards. As a whole, responses suggested that they felt the time is needed for parents to process information before the decision about screening is made; and that the decision be made during pregnancy (or, at the latest, around the time baby is born). About half of respondents suggested that verbal agreement to screening would be acceptable, but very few felt that consent could be assumed. The data did, however, clearly suggest that participants felt that health care providers and parents both have a responsibility to ensure that information is provided and understood.

Taken at face value, participants appeared to suggest that parents should have the responsibility to make a decision about NBS but that health care providers should be encouraging screening. About three-quarters seemed to agree that consent could be obtained by 'non-objection', and almost all suggested that a decision to decline screening should require a parent to sign a disclaimer. When asked directly about whether screening should be compulsory, responses were evenly split. Overall, these might suggest a group of respondents leaning strongly towards the positive value of NBS but wishing to retain parental authority.

In relation to how participants experienced the NBS decision for their youngest child, the scale scores did not signal serious loss of decisional control, although the responses to the specific items conveyed a sense that they did not have the autonomy they wished. Overall, respondents seemed somewhat actively engaged in information seeking and ready to seek out and make sense of information for themselves.

For the data on attitudes to healthcare and screening, the proportion of neutral responses was quite large for each item. However, generally, the responses leaned towards trusting the government as a provider of healthcare. There was a sense that the provision of a test like NBS as part of a routine service was an endorsement of its safety and benefits. The survey also included a set of items designed to provide insight into participants' socio-cultural worldview. As for the attitudes to healthcare, these were included to broaden out the types of personal attributes for inclusion in the latent class analysis. Overall, the mean scores for all three scales fell above the median, the highest for an orientation towards equity and fairness, and the lowest towards valuing freedom and personal autonomy. The discrete choice model for the overall sample was generally consistent with participants' attitudes towards newborn screening, as discerned from the descriptive analysis. Taken at face value, the key DCE results suggest that, overall, the participants would prefer to receive information about NBS somewhat later in pregnancy, that consent should not be assumed, and that their decision should be formally recorded.

The latent class analysis actually revealed that close to three-fifths of the participants, in fact, had no particular preferences in relation to the attributes that were examined and that the overall results were driven by two other (evenly split) groups with more distinct preferences: one where explicitness about the consent process was prioritized, and one where the timing of

information provision was prioritized. If valid, the limited analysis of the class membership characteristics suggests the consent oriented group generally feels competent to make NBS decisions and believes that parents should be given the authority and enough time to make them; and the information-oriented group is more oriented towards seeking out full information, and believes parents should be offered such information and time to make NBS decisions; possibly, this group may also interpret the offer of a screening test by a healthcare program as an endorsement of its safety and importance. Taken together, these groups could simply be considered as valuing consent as part of a conscious, informed decision-making process – providing the information that a person needs, giving them time to consider it, respecting their right to make the decision, and validating this with an explicit record.

The analysis also offers some limited insight into the personal characteristics associated with class membership, although no causal direction can be inferred. There were no strong demographic indicators, except that health professionals appeared most comfortable with an assumed consent approach. Individuals in the consent oriented group were possibly more likely to value individual autonomy generally. Those in the information oriented group were more likely to have reported that they were given insufficient time to make a screening decision for their youngest child.

The findings of this study are generally consistent with other studies about NBS decision-making or consent. Ulph, Dharni, Bennett, and Lavender (2019) found that assumed consent may be acceptable, but parents still wanted adequate information. They also noted that more explicit consent is preferred when aspects other than the screening test itself are considered, such as the practice of storing the bloodspot specimens for the longer term. Araia et al. (2012) suggest that parents prefer to have information about NBS prenatally rather than after the birth, consistent

with our findings. Fitzgerald et al. (2017) suggest that the provision of information about NBS during pregnancy can be inconsistent and that more structured or deliberate approaches are needed to address this. In an earlier report from this team's research program, based on qualitative interviews, Etchegary et al. (2016) observed differing expectations regarding NBS consent procedures between participants, with strong feelings on some that their consent should never be taken for granted.

Other studies have applied DCE approaches in the context of NBS, but none has examined the attributes of consent specifically. Miller et al. (2015) used a DCE to examine the public's opinion about NBS. Bombard et al. (2014) utilized a DCE to determine the perception of willingness and parental responsibility to participate in newborn screening using new genome technologies. The LC regression model was used by Carroll et al. (2013) to determine parents' preferences for Down's syndrome screening of their children.

4.1 Limitations of the study

A key limitation of this study is that participants were recruited via a non-probability based internet panel, so the sample cannot strictly be considered representative of the target population of all Canadian parents making NBS decisions. It is also not possible to tentatively judge selection bias by examing the response rate because, in practice, there was a clear-cut sampling frame available to the research team.

Overall, the survey participants were richer and had a higher proportion of people with undergraduate degrees or higher in education compared with the general population of the same age range (18 to 50 years old). This difference is consistent with the findings of Craig et al. (2013), which suggest that internet panel participants tend to have higher socioeconomic status than the general population. Also, participants in volunteer-based surveys often have strong

opinions about the subject, according to Statistics Canada (2017). Although the participant information consent form (see Appendix 5.4) masked the interest in consent specifically within a more general survey of NBS, it may still have attracted respondents with stronger than average opinions about the NBS based on their prior experiences. We also had no way of confirming the eligibility of the participants, and, arguably, a reward system used by a survey company might motivate ineligible panel members to participate inappropriately. It is not possible to validate the eligibility of the respondents, although this is not a potential problem that is confined to online surveys.

All self-completion surveys are subject to inaccuracies and bias in the responses they capture. The survey instrument itself was developed through a combination of validated instruments and items developed by the research team that was considered to have face validity (with some used in previous surveys) but had not been formally or fully validated. While there were reassuring consistencies between the preferences revealed in the discrete choice models for the different classes and the descriptive characteristics associated with the membership of these classes, we also noted inconsistent responses to items that were essentially measuring the same construct (e.g., recall of specific aspects of their own NBS experience, such as the earliest time information was received, how information is provided, and whether sufficient amount of information is provided, and opinions about whether NBS should be compulsory or whether parents should be allowed to say no to screening). There were some items that, in retrospect, would have been relevant and useful but were not included. The most obvious one was the jurisdiction within which the youngest child had been born or some proxy for this. In principle, the survey should also have confirmed the actual screening decision made by the parents so that a comparison could be made between those who accepted and those who declined. In practice,

the NBS uptake rate in Canada exceeds 99%, so that powering this study for a predefined comparison would have required either a much larger sample (for which the resources were not available) or a more sophisticated approach to the recruitment algorithm and implementation of the eligibility criteria in a quota sampling approach, including the consent form.

A limitation of using DCE to collect data was that the number of tasks required for achieving a 'full' balanced design depended on the number of attributes and the number of levels in each attribute (Kuhfeld, 2010). According to Kuhfeld (2010), the number of tasks required to achieve a 'full' balanced design may increase exponentially for every additional attribute introduced to the design. A total of 54 tasks would be required for a 'full' balanced design of DCE in this study. Participants may not have the patience to complete 54 choice tasks in a DCE. The DCE design in this study was not able to achieve a 'full' balance in order to reduce the burden of participants. Hence there might be bias in the DCE responses. Also, other attributes, such as the amount of information that should be provided (just enough vs. as much as possible) and the amount of pressure to consent (mandatory vs. encouraged vs. no pressure), were not included in this study.

Finally, a major limitation of the study is that it does not link the examination of preferences to any practical outcome for a consent or decision-making process for NBS. There is no agreed set of outcome measures in this area, although there is a general consensus that meeting parents' (or patients') preferences is desirable in itself. Although consent and informed decision making are not identical (Nicholls et al., 2019), the published literature on the latter suggests that interventions that promote shared decision making improve patients' knowledge of options, and agreement between patients' values and subsequent treatment or screening decisions (Coulter & Ellins, 2007).

Also, it was outside the scope of this thesis to examine the utility associated with different consent process models formally. Ideally, this should be done within a cost-utility analysis, which would have required a much larger study.

4.2 Value of the study and implications for policy and practice

Although published studies have used DCE to explore aspects of NBS, to our knowledge, this is the first that has specifically examined attributes of the consent process and used a technique such as LC regression to explore discrete variations in preference structures within a study sample. The results need to be validated in other studies, but they offer support to the assertion that preferences are not homogenous within populations as three classes of distinct preference patterns were discovered in our sample, and provide an insight into the information that might be gained through the thoughtful application of a method such as LC regression.

If the findings of this study are supported by other research, they can help informed discussion on possible improvements in NBS consent procedures in all Canadian provinces. Generally, the practice appears to be that parental consent is generally assumed, parents may not realize that their infant has even been screened, or they may only have had any kind of active information or discussion at the actual point of screening (Potter et al., 2014). Possibly, the majority of parents might be unconcerned about this, but a large minority seem not to be as informed and engaged as they would like, and some feel that their decision-making authority is undermined. DCE was crucial for discovering the values of attributes in the NBS consent procedure and how they traded off against each other despite its limitation in this study.

The data appear to suggest that there is no simple indicator of the likely preferences of an individual parent. However, there is a large body of evidence in other areas of healthcare that may suggest feasible and cost-effective interventions, such as decision aids, that could be

incorporated into the process in the prenatal period. Requiring that a consent decision be explicitly recorded in a patient's chart provides a simple indicator for later audit. Even simply requiring a parent's signature on the sample collection card would be a simple, quick intervention in itself, but anticipating the need to answer a parent's questions at that point would probably prompt consideration of improved information provision and discussion at an earlier stage.

4.3 Conclusion

In conclusion, this study assessed parents' past experiences of and attitudes towards NBS consent procedures and quantified their relative preferences for a set of attributes associated with them. The key findings related to – for a substantial minority of parents – the importance of timely and sufficient information provision and respect for parental authority in making the screening decision for their child. The current 'screening by assumed consent' model that appears to be widely followed in practice may not meet the preferences of this substantial minority. Interventions developed in other areas of healthcare might usefully be applied in the NBS setting.

Should the results of this project be confirmed by other studies, the next step would be to develop and evaluate interventions that support consent processes more aligned with the preferences identified. Such interventions might be educational – raising awareness of healthcare providers – but could also focus on re-engineering the 'standard' consent process to ensure that the 'default' moves closer to what many parents would prefer.

References

Andermann, A., Blancquaert, I., Beauchamp, S., & Déry, V. (2008). Revisiting Wilson and Jungner in the genomic age: a review of screening criteria over the past 40 years. *Bulletin of the World Health Organization*, *86*(4), 317–319. https://doi.org/10.2471/blt.07.050112

Araia, M. H., Wilson, B. J., Chakraborty, P., Gall, K., Honeywell, C., Milburn, J., Ramsay, T., & Potter, B. K. (2012). Factors associated with knowledge of and satisfaction with newborn screening education: a survey of mothers. *Genetics in Medicine : Official Journal of the American College of Medical Genetics*, *14*(12), 963–970. https://doi.org/10.1038/gim.2012.87

Department of Justice. (2019). *Article 12 of the Convention on the Rights of the Child and Children's Participatory Rights in Canada*. https://www.justice.gc.ca/eng/rp-pr/other-autre/article12/p3a.html

Becker, F., van El, C. G., Ibarreta, D., Zika, E., Hogarth, S., Borry, P., Cambon-Thomsen, A., Cassiman, J. J., Evers-Kiebooms, G., Hodgson, S., Janssens, A. C. J. W., Kaariainen, H., Krawczak, M., Kristoffersson, U., Lubinski, J., Patch, C., Penchaszadeh, V. B., Read, A., Rogowski, W., … Cornel, M. C. (2011). Genetic testing and common disorders in a public health framework: how to assess relevance and possibilities. Background Document to the ESHG recommendations on genetic testing and common disorders. *European Journal of Human Genetics : EJHG*, *19 Suppl 1*(Suppl 1), S6--S44. https://doi.org/10.1038/ejhg.2010.249

Berry, S. A., Brown, C., Grant, M., Greene, C. L., Jurecki, E., Koch, J., Moseley, K., Suter, R., van Calcar, S. C., Wiles, J., & Cederbaum, S. (2013). Newborn screening 50 years later: access issues faced by adults with PKU. *Genetics in Medicine : Official Journal of the American College of Medical Genetics*, *15*(8), 591–599. https://doi.org/10.1038/gim.2013.10

Bombard, Y., Miller, F. A., Hayeems, R. Z., Barg, C., Cressman, C., Carroll, J. C., Wilson, B. J., Little, J., Avard, D., Painter-Main, M., Allanson, J., Giguere, Y., & Chakraborty, P. (2014). Public views on participating in newborn screening using genome sequencing. *European Journal of Human Genetics : EJHG*, *22*(11), 1248–1254. https://doi.org/10.1038/ejhg.2014.22

Buri, J. R. (1991). Parental Authority Questionnaire. *Journal of Personality Assessment*, *57*(1), 110–119. https://doi.org/10.1207/s15327752jpa5701_13

Carpenter, K., Wittkowski, A., Hare, D. J., Medford, E., Rust, S., Jones, S. A., & Smith, D. M. (2018). Parenting a Child with Phenylketonuria (PKU): an Interpretative Phenomenological Analysis (IPA) of the Experience of Parents. *Journal of Genetic Counseling*, *27*(5), 1074–1086. https://doi.org/10.1007/s10897-018-0227-7

Carroll, F. E., Al-Janabi, H., Flynn, T., & Montgomery, A. A. (2013). Women and their partners' preferences for Down's syndrome screening tests: a discrete choice experiment. *Prenatal Diagnosis*, *33*(5), 449–456. https://doi.org/10.1002/pd.4086

https://doi.org/https://doi.org/10.1016/j.ymgme.2009.10.007

Cocanour, C. S. (2017). Informed consent---It's more than a signature on a piece of paper. *The American Journal of Surgery*, *214*(6), 993–997. https://doi.org/https://doi.org/10.1016/j.amjsurg.2017.09.015

Coulter, A., & Ellins, J. (2007). Effectiveness of strategies for informing, educating, and involving patients. *BMJ*, *335*(7609), 24. https://doi.org/10.1136/bmj.39246.581169.80

Cragun, D., DeBate, R. D., & Pal, T. (2015). Applying Public Health Screening Criteria: How Does Universal Newborn Screening Compare to Universal Tumor Screening for Lynch Syndrome in Adults with Colorectal Cancer? *Journal of Genetic Counseling*, *24*(3), 409–420. https://doi.org/10.1007/s10897-014-9769-5

DeLuca, J. M. (2018). Public Attitudes Toward Expanded Newborn Screening. *Journal of Pediatric Nursing*, *38*, e19--e23. https://doi.org/https://doi.org/10.1016/j.pedn.2017.10.002

Dhondt, J.-L. (2010). Expanded newborn screening: social and ethical issues. *Journal of Inherited Metabolic Disease*, *33*(S2), 211–217. https://doi.org/https://doi.org/10.1007/s10545-010-9138-y

DuBenske, L. L., Burke Beckjord, E., Hawkins, R. P., & Gustafson, D. H. (2009). Psychometric evaluation of the Health Information Orientation Scale: a brief measure for assessing health information engagement and apprehension. *Journal of Health Psychology*, *14*(6), 721–730. https://doi.org/10.1177/1359105309338892

Dziak, J. J., Lanza, S. T., and Tan, X. (2014). Effect Size, Statistical Power and Sample Size Requirements for the Bootstrap Likelihood Ratio Test in Latent Class Analysis. *Struct Equ Modeling*, *24*(*4*), 534–552. 10.1080/10705511.2014.919819

El-Hattab, A. W. (2015). Inborn Errors of Metabolism. *Clinics in Perinatology*, *42*(2), 413–439. https://doi.org/https://doi.org/10.1016/j.clp.2015.02.010

Etchegary, H., Nicholls, S. G., Tessier, L., Simmonds, C., Potter, B. K., Brehaut, J. C., Pullman, D., Hayeems, R., Zelenietz, S., Lamoureux, M., Milburn, J., Turner, L., Chakraborty, P., & Wilson, B. (2016). Consent for newborn screening: parents' and health-care professionals' experiences of consent in practice. *European Journal of Human Genetics : EJHG*, *24*(11), 1530–1534. https://doi.org/10.1038/ejhg.2016.55

Fitzgerald, C., Heery, E., Conneally, N., Linnane, B., George, S., & Fitzpatrick, P. (2017). An evaluation of pregnant women's knowledge and attitudes about newborn bloodspot screening. *Midwifery*, *45*, 21–27. https://doi.org/https://doi.org/10.1016/j.midw.2016.11.007

Ghanouni, A., Renzi, C., Meisel, S. F., & Waller, J. (2016). Common methods of measuring "informed choice" in screening participation: Challenges and future directions. *Preventive Medicine Reports*, *4*, 601–607. https://doi.org/10.1016/j.pmedr.2016.10.017

Gillon, R. (1994). Medical ethics: four principles plus attention to scope. *BMJ*, *309*(6948), 184. https://doi.org/10.1136/bmj.309.6948.184

Goossens, L. M. A., Utens, C. M. A., Smeenk, F. W. J. M., Donkers, B., van Schayck, O. C. P., & Rutten-van Mölken, M. P. M. H. (2014). Should I Stay or Should I Go Home? A Latent Class Analysis of a Discrete Choice Experiment on Hospital-At-Home. *Value in Health*, *17*(5), 588–596. https://doi.org/10.1016/j.jval.2014.05.004

Greene, W. H., & Hensher, D. A. (2003). A latent class model for discrete choice analysis: contrasts with mixed logit. *Transportation Research Part B: Methodological*, *37*(8), 681–698. https://doi.org/https://doi.org/10.1016/S0191-2615(02)00046-2

Grootendorst, D. C., Jager, K. J., Zoccali, C., & Dekker, F. W. (2009). Screening: why, when, and how. *Kidney International*, *76*(7), 694–699. https://doi.org/10.1038/ki.2009.232

Grosse, S D, Rogowski, W. H., Ross, L. F., Cornel, M. C., Dondorp, W. J., & Khoury, M. J. (2010). Population Screening for Genetic Disorders in the 21st Century: Evidence, Economics, and Ethics. *Public Health Genomics*, *13*(2), 106–115. https://doi.org/10.1159/000226594

Grosse, Scott D, Boyle, C. A., Kenneson, A., Khoury, M. J., & Wilfond, B. S. (2006). From Public Health Emergency to Public Health Service: The Implications of Evolving Criteria

for Newborn Screening Panels. *Pediatrics*, *117*(3), 923. https://doi.org/10.1542/peds.2005-0553
Gupta, S. (2016). Screening: Baby's first test. *Nature*, *537*(7621), S162--S164. https://doi.org/10.1038/537S162a
Hain, R. D. W. (2018). Voices of moral authority: parents, doctors and what will actually help. *Journal of Medical Ethics*, *44*(7), 458. https://doi.org/10.1136/medethics-2017-104705
Hall, A. E., Chowdhury, S., Pashayan, N., Hallowell, N., Pharoah, P., & Burton, H. (2014). What ethical and legal principles should guide the genotyping of children as part of a personalised screening programme for common cancer? *Journal of Medical Ethics*, *40*(3), 163. https://doi.org/10.1136/medethics-2012-101079
Hall, D. M., & Stewart-Brown, S. (1998). Screening in child health. *British Medical Bulletin*, *54*(4), 929–943. https://doi.org/10.1093/oxfordjournals.bmb.a011739
Halushka v. University of Saskatchewan. (1965).
Hammami, M. M., Al-Gaai, E. A., Al-Jawarneh, Y., Amer, H., Hammami, M. B., Eissa, A., & Qadire, M. Al. (2014). Patients'perceived purpose of clinical informed consent: Mill's individual autonomy model is preferred. *BMC Medical Ethics*, *15*(1), 2. https://doi.org/10.1186/1472-6939-15-2
Hauber, A. B., González, J. M., Groothuis-Oudshoorn, C. G. M., Prior, T., Marshall, D. A., Cunningham, C., IJzerman, M. J., & Bridges, J. F. P. (2016). Statistical Methods for the Analysis of Discrete Choice Experiments: A Report of the ISPOR Conjoint Analysis Good Research Practices Task Force. *Value in Health*, *19*(4), 300–315. https://doi.org/10.1016/j.jval.2016.04.004
Hayeems, R. Z., Miller, F. A., Bombard, Y., Avard, D., Carroll, J., Wilson, B., Little, J., Chakraborty, P., Bytautas, J., Giguere, Y., Allanson, J., & Axler, R. (2015). Expectations and values about expanded newborn screening: a public engagement study. *Health Expectations*, *18*(3), 419–429. https://doi.org/10.1111/hex.12047
Hays, R. D., Liu, H., & Kapteyn, A. (2015). Use of Internet panels to conduct surveys. *Behavior Research Methods*, *47*(3), 685–690. https://doi.org/10.3758/s13428-015-0617-9
Huber, M., Knottnerus, J. A., Green, L., Horst, H. van der, Jadad, A. R., Kromhout, D., Leonard, B., Lorig, K., Loureiro, M. I., Meer, J. W. M. van der, Schnabel, P., Smith, R., Weel, C. van, & Smid, H. (2011). How should we define health? *BMJ*, *343*, d4163. https://doi.org/10.1136/bmj.d4163
Jones, J. W., McCullough, L. B., & Richman, B. W. (2005). Informed Consent: It's Not Just Signing a Form. *Thoracic Surgery Clinics*, *15*(4), 451–460. https://doi.org/https://doi.org/10.1016/j.thorsurg.2005.06.001
Kahan, D. M., Braman, D., Gastil, J., Slovic, P., & Mertz, C. K. (2007). Culture and Identity-Protective Cognition: Explaining the White-Male Effect in Risk Perception. *Journal of Empirical Legal Studies*, *4*(3), 465–505. https://doi.org/10.1111/j.1740-1461.2007.00097.x
Kahan, D. M., Jenkins-Smith, H., & Braman, D. (2011). Cultural cognition of scientific consensus. *Journal of Risk Research*, *14*(2), 147–174. https://doi.org/10.1080/13669877.2010.511246
Katz, A. L., & Webb, S. A. (2016). Informed Consent in Decision-Making in Pediatric Practice. *Pediatrics*, *138*(2), e20161485. https://doi.org/10.1542/peds.2016-1485
Kaye, C. I. (2006). Newborn Screening Fact Sheets. *Pediatrics*, *118*(3), e934. https://doi.org/10.1542/peds.2006-1783
Khoury, M. J., McCabe, L. L., & McCabe, E. R. B. (2003). Population Screening in the Age of

Genomic Medicine. *New England Journal of Medicine*, *348*(1), 50–58. https://doi.org/10.1056/NEJMra013182
Kongsted, A., & Nielsen, A. M. (2017). Latent Class Analysis in health research. *Journal of Physiotherapy*, *63*(1), 55–58. https://doi.org/https://doi.org/10.1016/j.jphys.2016.05.018
Kuhfeld, W. F. (2010). *Discrete Choice*. http://support.sas.com/techsup/technote/mr2010f.pdf
Lachapelle, E., Montpetit, É., & Gauvin, J.-P. (2014). Public Perceptions of Expert Credibility on Policy Issues: The Role of Expert Framing and Political Worldviews. *Policy Studies Journal*, *42*(4), 674–697. https://doi.org/10.1111/psj.12073
Law, E. H., & Harrington, R. (2016). A Primer on Latent Class Analysis. *Value & Outcomes Spotlight*, 18–19.
Li, J. K. Y. (2020). Rethinking the Assessment of Decision-Making Capacity and Making Treatment-Related Decisions. *The Journal of Clinical Ethics*, *31*(1), 60–67.
Mak, C. M., Lee, H.-C. H., Chan, A. Y.-W., & Lam, C.-W. (2013). Inborn errors of metabolism and expanded newborn screening: review and update. *Critical Reviews in Clinical Laboratory Sciences*, *50*(6), 142–162. https://doi.org/10.3109/10408363.2013.847896
Miller, A. (2008). Screening: evidence and practice. *Bulletin of the World Health Organization*, *86*(4), 320. https://doi.org/10.2471/BLT.07.048744
Miller, F. A. (2009). The complex promise of newborn screening. *Indian J Med Ethics*, *6*(3), 142–148. https://doi.org/10.20529/IJME.2009.046
Miller, F. A., Hayeems, R. Z., Bombard, Y., Cressman, C., Barg, C. J., Carroll, J. C., Wilson, B. J., Little, J., Allanson, J., Chakraborty, P., Giguère, Y., & Regier, D. A. (2015). Public Perceptions of the Benefits and Risks of Newborn Screening. *Pediatrics*, *136*(2), e413. https://doi.org/10.1542/peds.2015-0518
Miller, V. A., Ittenbach, R. F., Harris, D., Reynolds, W. W., Beauchamp, T. L., Luce, M. F., & Nelson, R. M. (2011). The decision making control instrument to assess voluntary consent. *Medical Decision Making : An International Journal of the Society for Medical Decision Making*, *31*(5), 730–741. https://doi.org/10.1177/0272989X11398666
Neff, M. J. (2008). Informed Consent: What Is It? Who Can Give It? How Do We Improve It? *Respiratory Care*, *53*(10), 1337. http://rc.rcjournal.com/content/53/10/1337.abstract
Nicholls, S. G., Etchegary, H., Tessier, L., Simmonds, C., Potter, B. K., Brehaut, J. C., Pullman, D., Hayeems, R. Z., Zelenietz, S., Lamoureux, M., Milburn, J., Turner, L., Chakraborty, P., & Wilson, B. J. (2019). What is in a Name? Parent, Professional and Policy-Maker Conceptions of Consent-Related Language in the Context of Newborn Screening. *Public Health Ethics*, *12*(2), 158–175. https://doi.org/10.1093/phe/phz003
Pacifico, D., & Yoo, H. (2013). lclogit: A Stata command for fitting latent-class conditional logit models via the expectation-maximization algorithm. *Stata Journal*, *13*(3), 625–639. http://www.stata-journal.com/article.html?article=st0312
Petros, M. (2012). Revisiting the Wilson-Jungner criteria: how can supplemental criteria guide public health in the era of genetic screening? *Genetics in Medicine*, *14*(1), 129–134. https://doi.org/10.1038/gim.0b013e31823331d0
Potter, B. K., Etchegary, H., Nicholls, S. G., Wilson, B. J., Craigie, S. M., & Araia, M. H. (2015). Education and Parental Involvement in Decision-Making About Newborn Screening: Understanding Goals to Clarify Content. *Journal of Genetic Counseling*, *24*(3), 400–408. https://doi.org/10.1007/s10897-014-9780-x
Pullman, D. (2001). Subject comprehension, standards of information disclosure and potential liability in research. *Health Law J*, *9*, 113–127.

Reed Johnson, F., Lancsar, E., Marshall, D., Kilambi, V., Mühlbacher, A., Regier, D. A., Bresnahan, B. W., Kanninen, B., & Bridges, J. F. P. (2013). Constructing Experimental Designs for Discrete-Choice Experiments: Report of the ISPOR Conjoint Analysis Experimental Design Good Research Practices Task Force. *Value in Health*, *16*(1), 3–13. https://doi.org/10.1016/j.jval.2012.08.2223

Reibl v. Huges. (1980).

Robertson, G. (1991). Informed consent ten years later: the impact of Reibl v. Hughes. *The Canadian Bar Review*, *70*(3), 423–447. http://europepmc.org/abstract/MED/12211262

Ross, L. (2010). Mandatory versus Voluntary Consent for Newborn Screening? *Kennedy Institute of Ethics Journal*, *20*, 299–328.

Ross, L. F. (2006). Screening for conditions that do not meet the Wilson and Jungner criteria: The case of Duchenne muscular dystrophy. *American Journal of Medical Genetics Part A*, *140A*(8), 914–922. https://doi.org/10.1002/ajmg.a.31165

Ross, L. F. (2015). Ethical and Policy Issues in Newborn Screening of Children for Neurologic and Developmental Disorders. *Pediatric Clinics of North America*, *62*(3), 787–798. https://doi.org/https://doi.org/10.1016/j.pcl.2015.03.009

Saudubray, J.-M., & Garcia-Cazorla, À. (2018). Inborn Errors of Metabolism Overview: Pathophysiology, Manifestations, Evaluation, and Management. *Pediatric Clinics of North America*, *65*(2), 179–208. https://doi.org/https://doi.org/10.1016/j.pcl.2017.11.002

Shahpori, R., Hebert, M., Kushniruk, A., & Zuege, D. (2011). Telemedicine in the intensive care unit environment---A survey of the attitudes and perspectives of critical care clinicians. *Journal of Critical Care*, *26*(3), 328.e9--328.e15. https://doi.org/https://doi.org/10.1016/j.jcrc.2010.07.013

Statistics Canada. (2021). *Canadian Income Survey, 2019*. https://www150.statcan.gc.ca/n1/daily-quotidien/210323/dq210323a-eng.htm

Statistics Canada. (2017). *Non-probability sampling*. https://www150.statcan.gc.ca/n1/edu/power-pouvoir/ch13/nonprob/5214898-eng.htm#a2

Summers, J. (2007). *Principles of Healthcare Ethics*. Wiley.

Ulph, F., Dharni, N., Bennett, R., & Lavender, T. (2020). Consent for newborn screening: screening professionals' and parents'views. *Public Health*, *178*, 151–158. https://doi.org/https://doi.org/10.1016/j.puhe.2019.08.009

van der Burg, S., & Oerlemans, A. (2018). Fostering caring relationships: Suggestions to rethink liberal perspectives on the ethics of newborn screening. *Bioethics*, *32*(3), 171–183. https://doi.org/10.1111/bioe.12425

Vass, C. M., Georgsson, S., Ulph, F., & Payne, K. (2019). Preferences for aspects of antenatal and newborn screening: a systematic review. *BMC Pregnancy and Childbirth*, *19*(1), 131. https://doi.org/10.1186/s12884-019-2278-7

Vermont J. K. & Magidson, J. (2002). *Latet Class Analysis*.

von Tigerstrom, B. (2001). Informed Consent for Treatment: A Review of the Legal Requirements. *Journal SOGC*, *23*(10), 951–956. https://doi.org/10.1016/S0849-5831(16)30863-1

Vos, I. M. L., Schermer, M. H. N., & Bolt, I. L. L. E. (2018). Recent insights into decision-making and their implications for informed consent. *Journal of Medical Ethics*, *44*(11), 734. https://doi.org/10.1136/medethics-2018-104884

Wasim, M., Awan, F. R., Khan, H. N., Tawab, A., Iqbal, M., & Ayesha, H. (2018). Aminoacidopathies: Prevalence, Etiology, Screening, and Treatment Options. *Biochemical*

Genetics, *56*(1), 7–21. https://doi.org/10.1007/s10528-017-9825-6
Wogalter, M. (1999). On the adequacy of legal documents: factors that influence informed consent. *Ergonomics*, *42*, 593–613. https://doi.org/10.1080/001401399185504
Wright, S. J., Ulph, F., Dharni, N., & Payne, K. (2017). Eliciting Preferences for Information Provision in Newborn Bloodspot Screening Programs. *Value in Health*, *20*(4), 651–661. https://doi.org/10.1016/j.jval.2016.11.007

Appendices

5.1 Survey Instrumentation

Public experiences, attitudes, and preferences regarding consent procedures for newborn bloodspot screening

[For the REB, questions in this document are NOT check all that apply unless specified].

Newborn bloodspot screening (you may have heard this called newborn screening, the PKU test, or the heel prick) is a test that is usually done shortly after birth and involves taking a small sample of blood by pricking the baby's heel. At the moment, we know very little about how parents such as yourself experience newborn bloodspot screening.

In this survey we want to hear from you about your experiences of newborn bloodspot screening, and how you would like screening to be provided. We plan to use the information we collect to improve the way newborn bloodspot screening is provided.

Your participation in this survey is voluntary. We ask that you try to answer all of the questions. However, if you feel uncomfortable answering a particular question, you may leave it blank. The information that you provide will remain completely anonymous in all of our study reports.

Screening questionnaire

Before we begin, we have a few questions to confirm your eligibility for this study. All information will be treated in the strictest confidence and none will be shared with anyone outside the research team.

1. Are you the parent of one or more children aged 5 years or under?
   - [ ] Yes
   - [ ] No
2. Was your youngest child born in Canada?
   - [ ] Yes
   - [ ] No
3. Are you comfortable reading in English?
   - [ ] Yes
   - [ ] No
4. Are you at least 18 years of age?
   - [ ] Yes
   - [ ] No

Section 1: Your experiences of newborn screening

Here we ask you about your own experiences. Some of the questions are about what happened, for example who you saw, while others are about how you felt about things. We know that people will have had different experiences, and there are no right or wrong answers.

1. Do you recall your healthcare provider talking to you or providing information about newborn bloodspot screening?
   - [ ] No
   - [ ] Yes, the provider talked to me about newborn bloodspot screening
   - [ ] Yes, the provider gave me written information about newborn bloodspot screening
   - [ ] Yes, the provider gave me written information and we discussed newborn bloodspot screening

2. Do you recall reading the information provided about newborn bloodspot screening?
   - [ ] Yes
   - [ ] No

3. Please indicate when the information about newborn bloodspot screening was provided to you (check all that apply):
   - [ ] Early in pregnancy (first or second trimester)
   - [ ] Late in pregnancy (third trimester)
   - [ ] When the baby was born
   - [ ] When the sample was taken
   - [ ] After the sample was taken
   - [ ] I do not recall receiving information about newborn bloodspot screening

4. How do you feel about the AMOUNT of information you were provided about newborn screening?
   - [ ] I do not recall receiving information about newborn bloodspot screening
   - [ ] Too little
   - [ ] About right
   - [ ] Too much

5. Wherever you got your information, please indicate which pieces of information you felt you understood (please select all that apply):

[ ] The names of the conditions included in newborn bloodspot screening
[ ] How to prepare my baby for the sample being taken
[ ] How the sample would be taken
[ ] Whether my baby would suffer any pain or side effects when the sample is taken
[ ] The time it would take to receive results
[ ] How I would receive the results
[ ] Whether I could receive a result which suggested my child does not have a condition when in reality they did have one
[ ] Whether I could receive a result which suggested my child has a condition when in reality they did not have one
[ ] The effect that having one of the conditions would have on my child's health
[ ] How common or rare the conditions are
[ ] What could be done to treat my child should they have one of the conditions

6. Do you recall being asked to agree to have your child screened?

[ ] Yes
[ ] No

7. Do you recall when were you asked to agree to screening?

[ ] Early in pregnancy (first or second trimester)
[ ] Late in pregnancy (third trimester)
[ ] When the baby was born
[ ] When the sample was about to be taken
[ ] After the sample was taken
[ ] I can't remember when I was asked
[ ] I was not asked to agree to screening

Section 2: Your preferences for making decisions for yourself and others

We would like to know how you felt about deciding whether to have your baby screened or not.

8. For each of the statements below, please indicate the extent to which you feel it reflects your experience with the decision to screen or not screen your baby as part of newborn bloodspot screening.

	Strongly Disagree	Disagree	Somewhat Disagree	Somewhat Agree	Agree	Strongly Agree
I was powerless in the face of this decision	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
Someone took the decision away from me	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
I made the decision	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
I was passive in the face of this decision	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
The decision was inappropriately influenced by others	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
I was not in control of this decision	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
Others made the decision against my wishes	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
I was not the one to choose	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]
The decision was up to me	[ ]	[ ]	[ ]	[ ]	[ ]	[ ]

Section 3: Your preferences for how screening should be provided by the province

In the last section we asked you about your experiences. Now we would now like to ask you about how you feel newborn bloodspot screening SHOULD be <u>presented</u>.

9. For each statement please choose one option on the extent to which you agree or disagree.

	Agree	Somewhat Agree	Somewhat Disagree	Disagree
Parents should be strongly advised to have screening for their baby	[ ]	[ ]	[ ]	[ ]
Parents should have to actively choose to screen or not screen their baby	[ ]	[ ]	[ ]	[ ]
Screening should happen unless a parent objects	[ ]	[ ]	[ ]	[ ]
Parents should be made to sign a disclaimer if they choose not to have their baby screened	[ ]	[ ]	[ ]	[ ]
Parent should not be allowed to say no to screening	[ ]	[ ]	[ ]	[ ]

10. What would be your preferred role in making a decision about whether or not have newborn bloodspot screening for your baby?

[ ] I would prefer to make the decision about screening entirely by myself
[ ] I would prefer to make the final decision about screening after seriously considering my healthcare professional's opinion
[ ] I would prefer that my healthcare professional and I shared responsibility for deciding whether screening was best for my baby.
[ ] I would prefer that my healthcare professional made the final decision about whether to screen my baby, but seriously considered my opinion.
[ ] I would prefer to leave the decision regarding screening my baby to my healthcare professional

11. Do you think healthcare professionals have a responsibility to provide information to prospective parents about newborn bloodspot screening?

[ ] Yes
[ ] No (skip to question 13)

12. Please indicate when information should be provided to parents about newborn blood spot screening (please check all that apply):

[ ] Early pregnancy (first or second trimester)
[ ] Late pregnancy (third trimester)
[ ] When the baby is born
[ ] When the sample is taken
[ ] After the sample is taken

13. Please indicate which of the following types of information would be useful when learning about newborn bloodspot screening (select all that apply):

[ ] Discussion
[ ] Leaflet
[ ] The internet
[ ] Mobile/tablet app
[ ] Group session
[ ] Other (please specify)_____

14. Please indicate which of the following types of information would be MOST useful when learning about newborn bloodspot screening (please select one only):

[ ] Discussion
[ ] Leaflet
[ ] The internet
[ ] Mobile/tablet app
[ ] Group session
[ ] Other (please specify)_____

We would like to ask you about <u>when and how</u> the decision to screen SHOULD be made (this may be at a separate time to when information is provided).

15. When should a decision be made whether a baby will be screened or not?

[ ] Early in pregnancy (first or second trimester)
[ ] Late in pregnancy (third trimester)
[ ] When the baby is born
[ ] When the sample is about to be taken
[ ] Screening should be compulsory and a decision does not need to be made by the parent

16. Parents have a responsibility to review information materials provided to them about newborn bloodspot screening.

[ ] Yes
[ ] No

17. A parent's understanding of key pieces of information should be confirmed before screening proceeds.

[ ] Yes
[ ] No

18. How long should the time be between being given information and having to make a decision about screening?

[ ] No time, screening should not be subject to parental decision
[ ] Long enough to read the leaflet
[ ] Time to discuss in the moment
[ ] A week or two, enough time to look into things
[ ] More than two weeks

19. Screening may proceed if:

[ ] Agreed to verbally, no need for paper work
[ ] Agreed to verbally and noted by a healthcare professional
[ ] Recorded with written confirmation by parent
[ ] Agreement to proceed assumed if no objection made
[ ] Any of the above/does not really matter

Section 4: Your preferences for a screening service

Here we would like to explore your preferences for different ways that newborn bloodspot screening could be provided.

We focus on four aspects of the newborn bloodspot screening process. These are:

- The time at which information leaflets (or web links) about newborn bloodspot screening are provided. [“Best time to introduce”]
- The role of health professionals in supporting the decision to proceed with newborn bloodspot screening. [“Health professional role”]
- The reasonable assumption about the parent’s likely consent decision. [“Reasonable assumption about consent”]
- When it is necessary to record the consent decision in writing. [“Consent documentation”]

In this section, we will present different scenarios which reflect how these aspects can differ, and invite you to choose which you prefer.

[For the REB:

These are the complete scenario attributes and levels that will be presented to participants in various combinations:

Attribute	Levels
Best time to introduce information	Earlier in pregnancy (first or second trimester) so there's plenty of time to think about it
	Later in pregnancy so it's closer to the birth and more relevant to making the screening decision
	At the time of the actual heel prick when the sample is collected
Health professional role	To provide a clear recommendation about screening to parents
	To work with parents to come to a mutually agreeable decision
	To provide sufficient information for parents to make their own decision
Reasonable assumption about consent	Consent can reasonably be assumed unless the parent says otherwise
	Consent can never reasonably be assumed and should always be discussed with the parent
Consent documentation	It is seldom necessary to record the parents' consent decision in writing
	It is not necessary to record the parents' consent decision in writing unless screening is declined
	It is always necessary to record in writing the parents' consent decision

These will be combined into scenarios, and participants will be asked to select the scenario they prefer. Participants will be presented with 12 scenarios. Question 20 below provides an example of the question format.]

20. Considering your own experience of having a baby, which of the two scenarios below represents a better approach to providing newborn bloodspot screening?
REMEMBER: please choose which scenario you would prefer overall.

	Scenario A	**Scenario B**
Best time to introduce information	Earlier in pregnancy (first or second trimester) so there's plenty of time to think about it	Later in pregnancy so it's closer to the birth and more relevant to making the screening decision
Health professional role	To provide a clear recommendation about screening to parents	To provide sufficient information for parents to make their own decision.
Reasonable assumption about consent	Consent can reasonably be assumed unless the parent says otherwise.	Consent can reasonably be assumed unless the parent says otherwise.
Consent documentation	It is seldom necessary to record the parent consent decision in writing	It is always necessary to record in writing the parent consent decision
CHOOSE A or B	[ ]	[ ]

Section 5: Information about you

We would like more information about you, so we can examine how different people respond to our other questions. This will help us to identify if attitudes or experiences vary by individual characteristics.

All information will be treated in the strictest confidence and not be shared with anyone outside the research team.

21. Which age groups best fits you?

[ ] Less than 20 years
[ ] 20 to 24
[ ] 25 to 29
[ ] 30 to 34
[ ] 35 to 39
[ ] 40 to 44
[ ] 45 years or older

22. Which gender do you identify with?

[ ] Male
[ ] Female
[ ] You don't have an option that applies to me

22b. Please specify how you identify: _____

23. Are you an Aboriginal person, that is, North American Indian, Metis and/or Inuit?

[ ] Yes
[ ] No
[ ] Prefer not to answer

24. People living in Canada come from many different cultural and racial backgrounds. Are you (check all that apply):

[ ] White
[ ] Chinese
[ ] South Asian (e.g., East Indian, Pakistani, Sri Lankan)
[ ] Black
[ ] Filipino
[ ] Latin American
[ ] Southeast Asian (e.g., Cambodian, Indonesian, Laotian, Vietnamese)
[ ] Arab
[ ] West Asian (e.g., Afghan, Iranian)
[ ] Japanese
[ ] Korean
[ ] Other (please specify)

24b. If other, please specify: _____

25. Are you:
    [ ] Married
    [ ] Living common-law
    [ ] Widowed
    [ ] Separated
    [ ] Divorced
    [ ] Single-never married

26. What is the best estimate of your total household income?
    [ ] Less than $25,900
    [ ] $25,901 - $46,100
    [ ] $46,101 - $70,800
    [ ] $70,801 - $111,500
    [ ] More than $111,500
27. What is the highest degree, certificate or diploma you have obtained?
    [ ] No post-secondary degree, certificate or diploma
    [ ] Trade certificate or diploma from a vocational school or apprenticeship training
    [ ] Non-university certificate or diploma from a community college, CEGEP, school of nursing, etc.
    [ ] University certificate below bachelor's level
    [ ] Bachelor's degree
    [ ] University degree or certificate above bachelor's degree

28. Which of the following best describe where you live?
    [ ] Rural area
    [ ] Small city/town (less than 100,000 people
    [ ] Medium-sized city (100,000-499,999 people)
    [ ] Large city (500,000 or more people)
29. Language most commonly spoken at home?
    [ ] English
    [ ] French
    [ ] Other (please specify)_____
30. How many children do you have?
    [ ] One
    [ ] Two
    [ ] Three or more
31. Where was your youngest child born?
    [ ] In hospital
    [ ] In a birthing centre
    [ ] At home
    [ ] Other (please specify)_____
32. Are you a healthcare professional (e.g., nurse, doctor, dentist)?
    [ ] Yes
    [ ] No

We would like to ask you about how you prefer to make decisions involving your health.

33. Please mark how true each statement is for you:

	Not at all True	Slightly True	Moderately True	Very True	Very Much True
I like to gather as much information as I can before making a decision	[ ]	[ ]	[ ]	[ ]	[ ]
I like to review information multiple times before making a decision	[ ]	[ ]	[ ]	[ ]	[ ]
After I've made a decision, I continue to look for related information	[ ]	[ ]	[ ]	[ ]	[ ]
I like to make decisions quickly	[ ]	[ ]	[ ]	[ ]	[ ]
I have difficulty making sense of information from multiple sources	[ ]	[ ]	[ ]	[ ]	[ ]
I fear that I might find out something I don't want to know	[ ]	[ ]	[ ]	[ ]	[ ]
I feel overwhelmed by the amount of information available	[ ]	[ ]	[ ]	[ ]	[ ]
I think it's the doctor's job to deal with information, not mine	[ ]	[ ]	[ ]	[ ]	[ ]

34. Do you have spiritual/religious beliefs that influence your medical decisions?

[ ] Yes
[ ] No

35. Do you believe newborn bloodspot screening should be made compulsory?

[ ] Yes
[ ] No
[ ] No preference

36. People in our society often disagree about how far to let individuals go in making decisions for themselves. For each of the following statements, please indicate your level of agreement or disagreement:

	Strongly Agree	Moderately Agree	Moderately Disagree	Strongly Disagree
The government should put limits on the choices individual can make so they don't get in the way of what's good for society	[ ]	[ ]	[ ]	[ ]
The government should do more to advance society's goals, even if that means limiting the freedom and choices of individuals	[ ]	[ ]	[ ]	[ ]
Sometimes the government needs to make laws that keep people from hurting themselves	[ ]	[ ]	[ ]	[ ]
It's not the government's business to try to protect people from themselves	[ ]	[ ]	[ ]	[ ]
The government should stop telling people how to live their lives	[ ]	[ ]	[ ]	[ ]
The government interferes far too much in our everyday lives	[ ]	[ ]	[ ]	[ ]
We need to dramatically reduce inequalities between the rich and the poor, as well as between men and women	[ ]	[ ]	[ ]	[ ]
Our society would be better off if the distribution of wealth was more equal	[ ]	[ ]	[ ]	[ ]

37. Below are various healthcare services, organizations, and care providers that you might have contact with. You may have more confidence/trust in some services than others. Please indicate how much confidence/trust you have in them by choosing one response on each line:

	Strongly Disagree	Disagree	Neutral	Agree	Strongly Agree
The government will ensure a high quality health care system	[ ]	[ ]	[ ]	[ ]	[ ]
I feel like I have to double check everything the hospital does to my baby	[ ]	[ ]	[ ]	[ ]	[ ]
The hospital only provides screening tests that are important and safe	[ ]	[ ]	[ ]	[ ]	[ ]
Most medical researchers want to work on things that will make life better for the average person	[ ]	[ ]	[ ]	[ ]	[ ]
If the government has funded a health test or procedure, it's probably a worthwhile test to have	[ ]	[ ]	[ ]	[ ]	[ ]
The government wouldn't fund a health test or procedure if they were not sure of its benefits	[ ]	[ ]	[ ]	[ ]	[ ]

38. If you have any comments on the questionnaire or feel there is something we have not covered, please use the space below to let us know: _____

Thank you very much for taking the time to participate in this study. If you have any questions at all please do not hesitate to contact a study team member using the contact information provided below:

Principle Investigator, Dr. Beth Potter
Phone: 613-562-5800 ext. 8718
Email: bpotter@uottawa.ca

If you have questions about your right as a participant or about ethical issues related to this study, you can talk to someone who is not involved in the study by contacting the University of Ottawa Research Ethics Board:

Office of Research Ethics and Integrity
Taberet Hall
550 Cumberland St.
Room 154
Ottawa, ON, Canada
K1N 6N5

Phone: 613-562-5387
Fax: 613-562-5338
Email: ethics@uottawa.ca

5.2 Scales and Scoring Instructions

Appendix Table 5.1 Decision-Making Control Instrument (Miller et al. 2011)

Statements		Scores					
		Strongly Disagree	**Disagree**	**Somewhat Disagree**	**Somewhat Agree**	**Agree**	**Strongly Agree**
1	I was powerless in the face of this decision.	1	2	3	4	5	6
2	Someone took this decision away from me.	1	2	3	4	5	6
3	I made this decision.	1	2	3	4	5	6
4	I was passive in the face of this decision.	1	2	3	4	5	6
5	The decision about the protocol was inappropriately influenced by others.	1	2	3	4	5	6
6	I was not in control of this decision.	1	2	3	4	5	6
7	Others made this decision against my wishes.	1	2	3	4	5	6
8	I was not the one to choose.	1	2	3	4	5	6
9	The decision was up to me.	1	2	3	4	5	6

Scoring Instructions (Miller et al., 2011)

1. Reverse all scores except for statement 3 and 9
2. Sum of the converted scores for all statements to compute the total score
3. Subscales:
    a. Self-Control: sum of statements 3, 8, and 9
    b. Absense of Control: sum of statements 1, 4, and 6
    c. Others' Control: Sum of statements 2, 5, and 7

Appendix Table 5.2 Health Information Orientation (DuBenske et al., 2011)

Statements		Scores				
		Not true at all	**Slightly true**	**Moderately true**	**Very true**	**Very much true**
1	I like to gather as much information as I can before making a decision	0	1	2	3	4
2	I review information multiple times before making a decision	0	1	2	3	4
3	After I made a decision, I continue to look for related information	0	1	2	3	4
4	I like to make decisions quickly	0	1	2	3	4
5	I have difficulty make sense of information from multiple sources	0	1	2	3	4
6	I fear I might find out something I don't want to know	0	1	2	3	4
7	I feel overwhelmed by the amount of information available	0	1	2	3	4
8	I think it's the doctor's job to deal with information, not mine	0	1	2	3	4

Score Instrutctions (DuBenske et al., 2011)

Subscales:

1. Information Engegament: average score of statements 1-4
2. Information Apprehension: average score of statements 5-8

Appendix Table 5.3 Culture Worldview Scale (Lachapelle et al., 2014)

Statements		Scores			
		Strongly Agree	**Agree**	**Disagree**	**Strongly Disagree**
1	Government should put limits on the choices individuals can make so they don't get in the way of what's good for society	4	3	2	1
2	Government should do more to advance society's goals, even if that means limiting the freedom and choices of individuals	4	3	2	1
3	Sometimes government needs to make laws that keep people from hurting themselves	4	3	2	1
4	It's not the government's business to try to protect people from themselves	4	3	2	1
5	Government should stop telling people how to live their lives	4	3	2	1
6	Government interferes far too much in our everyday lives	4	3	2	1
7	We need to dramatically reduce inequalities between the rich and the poor, as well as between men and women	4	3	2	1
8	Our society would be better off if the distribution of wealth was more equal	4	3	2	1

Score Instruction (Lachapelle et al., 2014)

Subscales:

1. Hierarchism: average score of statements 1-3
2. Individualism: average score of statements 4-6
3. Egalitarianism: average score of statement 7 and 8

5.3 Data Sharing Agreement

Data Sharing Agreement ("Agreement")
Research Use of Data

BETWEEN:	AND
University of Ottawa 3042-800 King Edward Ottawa, Ontario K1N 6N5	**Memorial University of Newfoundland** Research Grant and Contract Services Bruneau Centre for Research and Innovation, 2nd Floor, IIC 2015 230 Elizabeth Avenue St. John's, NL Canada A1C 5S7
University of Ottawa Investigator: **Dr. Elizabeth K. Potter** having an academic appointment at uOttawa at 451 Smyth Rd., Ottawa, ON K1H 8M5 together with University of Ottawa = "**uOttawa**")	Memorial University of Newfoundland **Investigator:** **Dr. Brenda Wilson** (together with Memorial University of Newfoundland = "**MUN**")

Name of Study ("Study"): Public Experiences, attitudes, and preferences regarding consent procedures for newborn bloodspot screening.	
University of Ottawa **REB Study Number:** **H-12-19-5114**	Memorial University of Newfoundland **REB Study Number: 20210205-2020.072**

Data to be provided: De-identified data ("Data") per the REB approved Study Protocol, incorporated herein by reference.

This Agreement, effective as of the last date of signature below, is entered into between the parties to govern the transfer of Data between uOttawa and MUN to be used for the purposes of the Study in accordance with this Agreement. The party providing the Data is the "**PROVIDER**" and the party receiving the Data is the "**RECIPIENT.**"

PROVIDER will prepare and furnish to RECIPIENT the Data in accordance with applicable laws and Tri-Agency regulations, and specifically warrants that transfer of the Data by PROVIDER will be in compliance with REB approved subject informed consent forms ("ICFs") provided by the individuals from whom the Data were collected, or terms of an REB Waiver of Consent, as applicable. The Parties shall use a secure method of provision of the Data by PROVIDER to RECIPIENT. Data will not be transferred until each party's REB provides written approval for the Study.

RECIPIENT shall use the Data in compliance with all applicable laws; and shall specifically only use or disclose the Data for the conduct of the Study in accordance with the permitted uses of the Data specified in the applicable ICFs or REB Waiver of Consent, or otherwise as required by law. Notwithstanding any other provision of this Agreement, PROVIDER grants to RECIPIENT a worldwide, non-transferable, non-exclusive, royalty-free, irrevocable right and perpetual license to use all Data for educational, academic and research purposes.

It is the intention of the parties to publish results of this Study. RECIPIENT and PROVIDER shall have the right to use (1) the analyzed, de-identified data derived from the use of the Data, and (2) de-identified results arising out of analysis of the Data as part of a publication or presentation of the results of the Study. No personally identifying information shall be included in any publication or presentation of Study results. PROVIDER AND RECIPIENT shall coordinate publication of the results of the Study and appropriate authorship of any such publication shall be in accordance with academic standards and ICMJE guidelines.

RECIPIENT shall use appropriate safeguards to prevent any unauthorized use or disclosure of the Data and shall report to the PROVIDER any unauthorized use or disclosure of which RECIPIENT becomes aware, or of any breach of this Agreement. RECIPIENT shall not use the Data to identify or contact the individuals from whom such Data was collected. RECIPIENT shall securely destroy the Data as required by the Protocol or PROVIDER and provide a written

confirmation of the manner of destruction in a form acceptable to PROVIDER. PROVIDER may conduct audits, at its own expense, of the RECIPIENT concerning the maintenance of appropriate security safeguards to ensure compliance with this Agreement.

RECIPIENT shall give access to the Data only to its staff with a need to know for the purpose of conducting the Study, and who are bound by RECIPIENT to comply with the terms of this Agreement.

In the event that personal information or personal health information about a Study subject is inadvertently transferred to RECIPIENT or their respective employees or agents, RECIPIENT and their respective employees and agents shall not use or disclose such information and shall 1) immediately notify PROVIDER of receipt of such personal information or personal health information, 2) promptly destroy such personal information and personal health information in a secure fashion and 3) promptly certify such destruction in writing to PROVIDER. RECIPIENT shall take appropriate care in the disposal or destruction of the information to prevent unauthorized parties from gaining access to it. The parties shall make their employees and agents aware of the importance of maintaining the confidentiality of any collected or transferred personal health information or personal information. These obligations of confidentiality shall survive the expiration or earlier termination of this Agreement.

All notices, requests, directions or other communications ("**Notices**") required or permitted herein shall be in writing and will be delivered to the PROVIDER and RECIPIENT respectively at the addresses and to the individuals described below. In order for any such Notice to be effective, it will be delivered by email or by courier addressed to the PROVIDER or RECIPIENT for whom the Notices are intended at the above-mentioned address and will be deemed to have been received on the date of delivery, if delivered by courier, and on the next business day following the electronic confirmation of the successful transmission of the email, if sent by email. The address of the PROVIDER or RECIPIENT may be changed by notice in the manner set out in this section.

University of Ottawa:
For Contract Matters:
Director
Innovation Support Services
University of Ottawa
3042-800 King Edward Avenue
Ottawa, ON, K1N 6N5
Email: iss@uottawa.ca

For Scientific Matters:
Dr. Elizabeth Potter
University of Ottawa
451 Smyth Rd.
Ottawa, ON
K1H 8M5
beth.potter@uottawa.ca

Memorial University of Newfoundland:
For Contract Matters:
Mr. David Miller
Director, Research Grant and Contract Services
Memorial University of Newfoundland
230 Elizabeth Avenue
St. John's, NL
A1C 5S7
Email: rgcs@mun.ca

For Scientific Matters:
Dr. Brenda Wilson
Faculty of Medicine
Memorial University of Newfoundland
Health Science Centre H2843

300 Prince Philip Drive
St. John's, NL
A1B 3V6
Email: bwilson@mun.ca

Data are provided on an "as-is" basis and PROVIDER makes no representations or warranties, express or implied, with respect thereto. RECIPIENT accepts that there are no representations, warranties, conditions or liabilities expressed or implied herewith in relation to the Data by PROVIDER or its trustees, directors, officers, affiliates, investigators, students, employees, servants, authorized representatives or agents.

The Parties have requested that this Agreement and any related documents be drafted in the English language only. Les parties aux présentes ont exigé que la présente convention et tout document s'y rapportant soit rédigé en anglais seulement.

This Agreement may be signed in counterparts, and each counterpart may be delivered by facsimile or signed PDF by email. Each counterpart shall constitute an original, and when taken together, shall constitute one and the same instrument.

University of Ottawa	Memorial University of Newfoundland
Signature: I have authority to bin [redacted]	Signature: I have autho [redacted]
Date:	Date:
Name & Title: Brian Julien, Assistant Director Innovation Support Services	Name & Title: David Miller Director, Research Grant and Contract Services
University of Ottawa Investigator	Memorial University of Newfoundland **Investigator**
Signature: [redacted]	I have read and acknowledge the terms and conditions of this agreement and hereby agree to act in accordance with them Signature: [redacted]
Date: August 13, 2020	Date:
Dr. Elizabeth K. Potter	**Dr. Brenda Wilson**

5.4 Survey Participation Consent Form

PARTICIPANT INFORMED CONSENT FORM

[For the REB: panelists will be presented with this form after following the survey link provided by Dynata. This form will immediately precede the beginning of the survey]

Study Title: Public experiences, attitudes, and preferences regarding consent procedures for newborn bloodspot screening

Thank you for considering participation in this study. Please read the participation information provided below and in the following pages.

Principal Investigator (PI): Dr. Beth Potter, School of Epidemiology and Public Health, University of Ottawa, 613-562-5800 ext. 8718, bpotter@uottawa.ca

Funder: Canadian Institutes for Health Research (CIHR)

INTRODUCTION

You are being invited to take part in a research study. Before agreeing to participate in this study it is important you read and understand the following explanation of the proposed study. Please feel free to contact the research team to ask any questions. If you do not wish to participate you can simply ignore the invitation you have received.

BACKGROUND

What is the purpose of this study?

This study is about a screening program that is routinely offered to parents of newborn children. The newborn screening process involves taking a small sample of blood from the heel of the newborn and tested for a number of serious conditions that are not always obvious at birth. Identifying these conditions early on provides information that might prevent health problems such as developmental delay. This study aims to improve newborn screening by exploring parents' experiences with being offered screening, and their attitudes to how the process works.

How is the study designed?

This study is an online survey with five sections. Section 1 asks about your experiences with newborn screening when you had your child. Sections 2 asks about your preference for some aspects of the consent process, such as when you should receive information about newborn screening. Section 3 asks you about you preferences for how screening should be provided by the province. Section 4 asks you to choose what you believe an ideal consent process should be. Section 5 asks about your demographic information (this information will only be used to see if people with different characteristics have different views).

Who can participate?

You are invited to participate in this research study because you are a parent of a child born in Canada between the ages of 0-5 years. We are looking for parents to complete the survey. Because of this you must be able to read and understand English, the language of the survey. These are the only qualifications to participate in this study. If you choose to participate, a screening questionnaire will be administered before the beginning of the survey to confirm your eligibility.

How many people will take part in this study?

It is anticipated that approximately 700 participants from across Canada will take part in this study. This study should take one year to complete from the time of the first survey until the submission of the final manuscript.

MORE INFORMATION ABOUT THIS STUDY

What is expected of me?

As part of this study, you will be asked to complete a one-time online survey. It will take approximately 20-30 minutes to complete. If you do not wish to answer a question, you can choose not to.

How long will I be involved in the study?

Your participation in the study will end once you complete the survey. No further action is required from you.

What are the risks or harm of participating in this study?

There are no serious risks involved in this study. Although we are taking all necessary steps to keep all of the information you provide confidential, we cannot guarantee absolute security as the data are transmitted electronically.

What is the cost to participants?

Participation in this study will not involve any additional costs to you except for the time required to complete the survey.

What are the benefits of participating in this study?

You will not receive a direct health benefit from your participation in this study. This results of this research may help to improve service delivery for newborn bloodspot screening for future parents.

Are participants paid to be in this study?

You will be awarded points for your participation in this study, which you can redeem for products of your choice as per your original contract with Dynata.

How will participant information be kept confidential?

Your information will be kept strictly confidential. The study team will not have access to nor will we be collecting any personal health and identifying information from you in the survey. All your response data will be stored in a secure server provided by Sawtooth Software and/or in password protected drives and computers at the Faculty of Medicine, University of Ottawa or Faculty of Medicine, Memorial University of Newfoundland. Research records will be kept for 7 years, as required by the University of Ottawa Research Ethics Board. At the end of the storage time, all electronic records will be securely deleted.

For audit purposes only, original study files may be reviewed under the supervision of Dr. Potter's staff by representatives from the University of Ottawa, Memorial University of Newfoundland, or Sawtooth Software. The records received by these organization will not contain any identifying information. The Sawtooth Software server is housed in the United States and is therefore subject to the Patriot Act of the United States of America which allows American authorities to access your data. However, we are not collecting any identifying information and there is a minimal risk to participant confidentiality.

It is expected that the information collected during this study will be used in analyses and will be published/presented to the scientific community at meetings and in journals. You will not be identified in any publications or presentations resulting from this study.

In order to minimize the risk of security breaches and to help ensure your confidentiality, we recommend that you use standard safety measure such as signing out of your account, closing your browser and locking your screen or device when you are no long using them or when you have completed the study.

If I choose to participate can I withdraw later?

You do not have to reach the end of the survey and are free to withdraw at any time, but the data collected up until the point of withdrawal will be used in study analyses and reports as there will be no way for us to link your data to your identity. If you withdraw, you will not be identified from your responses. You may wish to review your contract with Dynata regarding their compensation policy for incomplete surveys. You will not be penalized by Dynata for choosing to not complete this survey.

Who do I contact for questions?

If you have any questions about taking part in this study you can talk to the person who is in charge of the study or a member of the research team using the contact information below:

Principal Investigator, Dr. Beth Potter

Phone: 613-562-5800 ext. 8718
Email: bpotter@uottawa.ca

If you have questions about your right as a participant or about ethical issues related to this study, you can talk to someone who is not involved in the study by contacting the University of Ottawa Research Ethics Board:

Office of Research Ethics and Integrity
Taberet Hall
550 Cumberland St.
Room 154
Ottawa, ON, Canada
K1N 6N5

Phone: 613-562-5387
Fax: 613-562-5338
Email: ethics@uottawa.ca

CHOOSING TO CONTINUE INDICATES:

- All of my questions regarding the study have been answered.
- I understand the previously presented information and instructions.
- I do not give up any of my legal rights by reading the previously presented information and instructions nor by participating in the study outlined previously.
- I agree to take part in this study.

5.5 HREB Approval Letter

Research Ethics Office
Suite 200, Eastern Trust Building
95 Bonaventure Avenue
St. John's, NL
A1B 2X5

May 27, 2020

Dear Dr Wilson:

Researcher Portal File # [redacted]
Reference # [redacted]

RE: Public experiences, attitudes, and preferences regarding consent procedures for newborn bloodspot screening

Your application was reviewed by a subcommittee under the direction of the HREB and your response was reviewed by the Chair and the following decision was rendered:

X	Approval
	Approval subject to changes
	Rejection

Ethics approval is granted for one year effective May 27, 2020. This ethics approval will be reported to the board at the next scheduled HREB meeting.

Please note this approval is deferred until the public health crisis related to COVID-19 has abated. No new studies involving face-to-face contact may be initiated at this time. Once this crisis has abated you will receive a communication with a revised letter. Do not begin any recruitment, consent processes, or study interventions until you receive your revised letter. If you have any questions please contact the Ethics Officer at ethicsofficer@hrea.ca. Thank you for your patience.

This is to confirm that the HREB reviewed and approved or acknowledged the following documents (as indicated):

- Application, approved

- Research proposal, approved
- Variable list, approved
- Budget, approved
- Participant consent form, approved
- University of Ottawa Certificate of Ethics Approval, acknowledged

Please note the following:

- This ethics approval will lapse on May 27, 2021. It is your responsibility to ensure that the Ethics Renewal form is submitted prior to the renewal date.
- This is your ethics approval only. Organizational approval may also be required. It is your responsibility to seek the necessary organizational approvals.
- Modifications of the study are not permitted without prior approval from the HREB. Request for modification to the study must be outlined on the relevant Event Form available on the Researcher Portal website.
- Though this research has received HREB approval, you are responsible for the ethical conduct of this research.
- If you have any questions please contact info@hrea.ca or 709 777 6974.

The HREB operates according to the Tri-Council Policy Statement: Ethical Conduct for Research Involving Humans (TCPS2), ICH Guidance E6: Good Clinical Practice Guidelines (GCP), the Health Research Ethics Authority Act (HREA Act) and applicable laws and regulations.

We wish you every success with your study.

[redacted]

You Have Received Ethics Approval, Now What?: HREB Reporting Requirements

Once a study has received ethics approval from the Health Research Ethics Board (HREB), there are still associated reporting requirements. In the conduct of approved research researchers are required to report to the HREB, in a timely manner, proposed changes from approved research that affect participants at any stage of the process. This includes, but is not limited to, changes to the consent form, changes to the tasks or interventions involved in the research, or changes to measures to protect privacy and confidentiality.

Any substantive change to the research should not be implemented prior to documented approval by the HREB, except when necessary to eliminate an immediate risk(s) to the participants. Below are examples of post approval documentation that must be submitted to the HREB:

Amendments

Any proposed change in the conduct of a study must be submitted to the HREB, and approved, before the change may be implemented. Such changes might include modification of recruitment procedures, inclusion or exclusion criteria, revised sample size, addition or deletion of study sites, changes to an intervention, consent forms, questionnaires or scripts, etc. If there are changes in project team members or changes to funding source(s)/sponsor(s), there are specific forms to complete to report this to the HREB.

Adverse Events

Serious and unanticipated adverse events that occur within Newfoundland and Labrador are required to be reported to the HREB. Such events may occur in both clinical trials and in other types of research, e.g. collapse during a rehabilitation program, emotional breakdown requiring follow up care during an interview, or breach of privacy during correspondence. Serious adverse events that are fatal or life-threatening are required to be reported to the HREB as soon as the research team is aware of the event.

Protocol Deviations

Deviations from an approved study protocol must be reported to the HREB. Changes that eliminate immediate hazards to participants do not require prior approval, but must be reported soon as reasonably possible.

Safety Reports

Safety reports providing information on all serious adverse events (SAEs) occurring in a clinical trial must be provided by the sponsor to the HREB, normally on a three or six monthly basis (i.e. in accordance with the specified reporting timelines that were outlined in the approved ethics application).

Investigator Brochure (IB) and Product Monograph (PM)

Throughout the course of a clinical trial, changes may be implemented to study documents. All revisions to approved study documents must be submitted to the HREB to ensure the record is up to date. If the revisions include new risk or safety information there may be a requirement to notify research participants.

Ethics Renewal/Study Closure

Ethics approval lasts for one year. Ethics renewal is required annually, on the anniversary of the date of the HREB notification of approval. Once data collection is no longer ongoing, a study closure form is required to be submitted to the HREB for the study to remain active or to be closed in good standing.

5.6 Ottawa REB Approval Letter

17/03/2020

Université d'Ottawa
Bureau d'éthique et d'intégrité de la recherche

University of Ottawa
Office of Research Ethics and Integrity

CERTIFICAT D'APPROBATION ÉTHIQUE I CERTIFICATE OF ETHICS APPROVAL

Numéro du dossier / Ethics File Number	[redacted]
Titre du projet / Project Title	Public Experiences, Attitudes, and Preferences Regarding Consent Procedures for Newborn Bloodspot Screening
Type de projet / Project Type	Recherche de professeur / Professor's research project
Statut du projet / Project Status	Approuvé / Approved
Date d'approbation (jj/mm/aaaa) / Approval Date (dd/mm/yyyy)	17/03/2020
Date d'expiration (jj/mm/aaaa) / Expiry Date (dd/mm/yyyy)	15/12/2020

Équipe de recherche / Research Team

Chercheur / Researcher	Affiliation	Role
Elizabeth POTTER	Département d'épidémiologie et santé publique / Department of Epidemiology and Public Health	Chercheur Principal / Principal Investigator
[redacted]	[redacted]	Coordonnateur de recherche / Research Coordinator

Conditions spéciales ou commentaires / Special conditions or comments

550, rue Cumberland, pièce 154 Ottawa (Ontario) K1N 6N5 Canada
550 Cumberland Street, Room 154 Ottawa, Ontario K1N 6N5 Canada

613-562-5387 • 613-562-5338 • ethique@uOttawa.ca / ethics@uOttawa.ca
www.recherche.uottawa.ca/deontologie I www.recherche.uottawa.ca/ethics

Université d'Ottawa
Bureau d'éthique et d'intégrité de la recherche

University of Ottawa
Office of Research Ethics and Integrity

Le Comité d'éthique de la recherche (CÉR) de l'Université d'Ottawa, opérant conformément à l'*Énoncé de politique des Trois conseils* (2014) et toutes autres lois et tous règlements applicables, a examiné et approuvé la demande d'éthique du projet de recherche ci-nommé.

L'approbation est valide pour la durée indiquée plus haut et est sujette aux conditions énumérées dans la section intitulée "Conditions Spéciales ou Commentaires". Le formulaire « Renouvellement ou Fermeture de Projet » doit être complété quatre semaines avant la date d'échéance indiquée ci-haut afin de demander un renouvellement de cette approbation éthique ou afin de fermer le dossier.

Toutes modifications apportées au projet doivent être approuvées par le CÉR avant leur mise en place, sauf si le participant doit être retiré en raison d'un danger immédiat ou s'il s'agit d'un changement ayant trait à des éléments administratifs ou logistiques du projet. Les chercheurs doivent aviser le CÉR dans les plus brefs délais de tout changement pouvant augmenter le niveau de risque aux participants ou pouvant affecter considérablement le déroulement du projet, rapporter tout évènement imprévu ou indésirable et soumettre toute nouvelle information pouvant nuire à la conduite du projet ou à la sécurité des participants.

The University of Ottawa Research Ethics Board, which operates in accordance with the *Tri-Council Policy Statement* (2014) and other applicable laws and regulations, has examined and approved the ethics application for the above-named research project.

Ethics approval is valid for the period indicated above and is subject to the conditions listed in the section entitled "Special Conditions or Comments". The "Renewal/Project Closure" form must be completedfour weeks before the above-referenced expiry date to request a renewal of this ethics approval or closure of the file.

Any changes made to the project must be approved by the REB before being implemented, except when necessary to remove participants from immediate endangerment or when the modification(s) only pertain to administrative or logistical components of the project. Investigators must also promptly alert the REB of any changes that increase the risk to participant(s), any changes that considerably affect the conduct of the project, all unanticipated and harmful events that occur, and new information that may negatively affect the conduct of the project or the safety of the participant(s).

Marc Alain BONENFANT
Coordonnateur de l'éthique / Ethics Coordinator
Pour/For **Daniel LAGAREC** Président(e) du/ Chair of the **Comité d'éthique de la recherche en sciences de la santé et sciences / Health Sciences and Sciences Research Ethics Board**

550, rue Cumberland, pièce 154
Ottawa (Ontario) K1N 6N5 Canada

550 Cumberland Street, Room 154
Ottawa, Ontario K1N 6N5 Canada

613-562-5387 • 613-562-5338 • ethique@uOttawa.ca / ethics@uOttawa.ca
www.recherche.uottawa.ca/deontologie I www.recherche.uottawa.ca/ethics

5.7 Additional Tables

Appendix Table 5.4: Views on the extent to which screening should be voluntary or compulsory, by class

Statement		N (%) 1=agree, 4=disagree				
	Class	N	1	2	3	4
Parents should have actively choese to screen or not to screen or not to screen their baby	Consent	148	72 (48.6)	58 (39.2)	10 (6.8)	8 (5.4)
	Information	147	67 (45.6)	54 (36.7)	16 (10.9)	10 (6.8)
	Flexible	418	150 (35.9)	185 (44.3)	62 (14.8)	21 (5.0)
Parents should be strongly advised to have screening for their baby	Consent	148	80 (54.0)	51 (34.5)	13 (8.8)	4 (2.7)
	Information	147	92 (62.6)	45 (30.6)	8 (5.4)	2 (1.4)
	Flexible	418	223 (53.4)	145 (34.7)	44 (10.5)	6 (1.4)
Screening should happen unless a parent objects	Consent	148	43 (29.1)	52 (35.1)	42 (28.4)	11 (7.4)
	Information	147	60 (40.8)	53 (36.0)	27 (18.4)	7 (4.8)
	Flexible	417	162 (38.8)	165 (39.6)	67 (16.1)	23 (5.5)
Parents should be made to sign a disclaimer if they choose not to have their baby screened	Consent	148	83 (56.1)	45 (30.4)	15 (10.1)	5 (3.4)
	Information	147	86 (58.5)	39 (26.5)	17 (11.6)	5 (3.4)
	Flexible	417	185 (44.4)	176 (42.2)	37 (8.9)	19 (4.5)
Parents should not be allowed to say no to screening	Consent	148	19 (12.8)	38 (25.7)	44 (29.7)	47 (31.8)
	Information	147	14 (9.5)	39 (26.5)	59 (40.2)	35 (23.8)
	Flexible	417	98 (23.5)	144 (34.5)	114 (27.4)	61 (14.6)

Appendix Table 5.5a: Recall of NBS information provision for youngest child, by class

Aspect	Categories	N	Consent	Information	Flexible
How information was provided by healthcare providers N (%)	Not provided or cannot remember Provided verbally only Provided in written format only Provided both verbally and in written format	$N_1 = 148$ $N_2 = 147$ $N_3 = 418$	39 (26.3) 63 (42.6) 21 (14.2) 25 (16.9)	55 (37.4) 54 (36.7) 17 (11.6) 21 (14.3)	115 (27.5) 162 (38.8) 71 (17.0) 70 (16.7)
Earliest time when information was provided N (%)	Early in pregnancy Late in pregnancy When the baby was born When sample was taken After sample was taken Cannot remember or no information received	$N_1 = 148$ $N_2 = 147$ $N_3 = 419$	27 (18.2) 27 (18.2) 38 (25.7) 14 (9.5) 2 (1.4) 40 (27.0)	15 (10.2) 27 (18.4) 38 (25.8) 17 (11.6) 4 (2.7) 46 (31.3)	107 (25.5) 78 (18.6) 93 (22.2) 38 (9.1) 15 (3.6) 88 (21.0)
Sufficiency of information provided N (%)	No information received Too little Just right Too much	$N_1 = 148$ $N_2 = 147$ $N_3 = 418$	36 (24.3) 40 (27.0) 67 (45.3) 5 (3.4)	45 (30.6) 45 (30.6) 55 (37.4) 2 (1.4)	88 (21.1) 95 (22.7) 219 (52.4) 16 (3.8)
Understanding of information content N (%)					
Name of conditions screened for		$N_1 = 148$ $N_2 = 147$ $N_3 = 420$	49 (33.1)	46 (31.3)	141 (33.6)
How to prepare baby for test			41 (27.7)	38 (25.9)	156 (37.1)
How the sample would be taken			81 (54.7)	87 (59.2)	196 (46.8)
Possible pain or side effects when the sample is taken			62 (41.9)	47 (32.0)	145 (34.5)
Time to receive results			49 (33.1)	37 (25.2)	134 (31.9)
How the results would be communicated			45 (30.4)	41 (27.9)	115 (27.4)
Possibility of false negative results			19 (12.8)	15 (10.2)	80 (19.1)
Possibility of false positive results			19 (12.8)	8 (5.4)	60 (14.3)
Health effects of conditions being screened for			27 (18.2)	25 (17.0)	78 (18.6)
Rarity of conditions being screened for			34 (23.0)	35 (23.8)	68 (16.2)
Treatment options for conditions being screened for			23 (15.5)	21 (14.3)	52 (12.4)

Appendix Table 5.5b: Recall of consent timing, by class

Recall of when consent was sought	N	Consent	Information	Flexible
Early in pregnancy	$N_1 = 147$ $N_2 = 147$ $N_3 = 419$	17 (11.6)	8 (5.4)	65 (15.5)
Late in pregnancy		14 (9.5)	18 (12.2)	69 (16.5)
When the baby was born		38 (25.8)	36 (24.5)	102 (24.3)
When the sample was taken		22 (15.0)	21 (14.3)	41 (9.8)
After the sample was taken		1 (0.7)	1 (0.7)	19 (4.5)
Does not recall when was consent asked		26 (17.7)	23 (15.7)	53 (12.6)
Never asked		29 (19.7)	40 (27.2)	70 (16.7)

Appendix Table 5.6a: Views on NBS information provision for parents, by class

Aspect	Categories	N	Consent	Information	Flexible
Earliest time when NBS information should be provided to parents N (%)	Early in pregnancy Late in pregnancy When the baby is born When sample about to be taken After the sample is taken	$N_1 = 148$ $N_2 = 147$ $N_3 = 419$	77 (52.0) 55 (37.2) 9 (6.1) 5 (3.4) 2 (1.3)	73 (49.7) 60 (40.8) 12 (8.2) 2 (1.4) 0 (0.0)	173 (41.3) 157 (37.5) 69 (16.4) 15 (3.6) 5 (1.2)
Ways of providing information that would be useful N (%)[1]	Through discussion Leaflet Internet Mobile/tablet app Group session	$N_1 = 148$ $N_2 = 147$ $N_3 = 420$	124 (83.8) 103 (69.6) 65 (43.9) 45 (30.4) 29 (19.6)	127 (86.4) 109 (74.2) 57 (38.8) 28 (19.1) 16 (10.9)	274 (65.2) 223 (53.1) 199 (47.4) 112 (26.7) 45 (11.7)
The single most useful way of providing information N (%)	Through discussion Leaflet Internet Mobile/tablet app Group session	$N_1 = 146$ $N_2 = 147$ $N_3 = 417$	78 (53.4) 34 (23.3) 17 (11.7) 11 (7.5) 6 (4.1)	79 (53.7) 44 (29.9) 16 (10.9) 3 (2.1) 5 (3.4)	166 (39.8) 98 (23.5) 101 (24.2) 38 (9.1) 14 (3.4)
When the screening decision should be made N (%)	Early in pregnancy Late in pregnancy When the baby is born When the sample is about to be taken Screening should be compulsory	$N_1 = 148$ $N_2 = 146$ $N_3 = 420$	39 (26.4) 52 (35.1) 48 (32.4) 6 (4.1) 3 (2.0)	30 (20.6) 60 (41.1) 46 (31.5) 6 (4.1) 4 (2.7)	114 (27.2) 126 (30.0) 111 (26.4) 37 (8.8) 31 (7.6)
Amount of time that should be given for the parent to decide N (%)	Screening not subject to parental decision Long enough to read the leaflet Time to discuss in the moment A week or two More than two weeks	$N_1 = 147$ $N_2 = 147$ $N_3 = 419$	2 (1.4) 43 (29.2) 30 (20.4) 57 (38.8) 15 (10.2)	9 (6.1) 19 (12.9) 33 (22.5) 66 (44.9) 20 (13.6)	50 (11.9) 124 (29.6) 143 (34.2) 81 (19.3) 21 (5.0)
Condition for allowing screening to proceed N (%)	Verbal agreement alone Verbal agreement, recorded by provider Written agreement by parent If no parental objection, may be assumed Does not really matter	$N_1 = 147$ $N_2 = 146$ $N_3 = 418$	5 (3.4) 54 (36.7) 84 (57.1) 3 (2.1) 1 (0.7)	15 (10.3) 53 (36.3) 58 (39.7) 8 (5.5) 12 (8.2)	72 (17.2) 152 (36.4) 137 (32.8) 27 (6.4) 30 (7.2)

[1] Multiple responses

Appendix Table 5.6b: Views on ensuring that information is provided and understood, by class

Responsibility	Class	N	Response
Healthcare professionals have a responsibility for providing information about NBS (agree)	Consent Information Flexible	148 147 417	144 (97.3) 145 (98.6) 394 (94.5)
Parents have a responsibility to review information materials provided to them about NBS (agree)	Consent Information Flexible	147 147 415	140 (95.2) 146 (99.3) 372 (89.4)
A parent's understanding of key pieces of information should be confirmed before screening proceeds (agree)	Consent Information Flexible	148 145 417	146 (98.7) 141 (97.2) 371 (89.0)

Appendix Table 5.7: Feelings about experience of NBS decision making (DMCI (Miller et al., 2011)), scale and sub-scale scores, by class

Scale	Class	N	Mean	SD[1]	Min	Max
Total Score	Consent	144	38.2	9.72	9	54
	Information	141	37.3	10.7		
	Flexible	409	34.3	9.39		
Self-control	Consent	147	12.6	3.84	3	18
	Information	143	11.8	4.25		
	Flexible	415	11.8	3.41		
Absence of Control	Consent	147	12.1	3.82	3	18
	Information	146	11.9	4.08		
	Flexible	414	10.8	3.60		
Others' Control	Consent	145	13.4	3.82	3	18
	Information	143	13.5	3.79		
	Flexible	414	11.7	4.05		

[1] Standard Deviation

Appendix Table 5.8: Information to support decision-making, in general (Health Information Orientation Scale (DuBenske et al., 2009)), sub-scale scores, by class

Scale	Class	N	Mean	SD[1]	Min	Max
Information Engagement	Consent	146	2.41	0.61	0	4
	Information	146	2.38	0.71		
	Flexible	420	2.44	0.75		
Information Apprehension	Consent	146	1.35	0.93	0	4
	Information	146	1.09	0.91		
	Flexible	420	1.79	1.00		

[1] Standard Deviation

Appendix Table 5.9: Views on trustworthiness of healthcare, by class

Statements	Class	N	Responses (Percentage)		
			Disagree	Neutral	Agree
The government will ensure a high quality health care system	Consent	148	28 (18.9)	37 (25.0)	83 (56.1)
	Information	147	19 (12.9)	42 (28.6)	86 (58.5)
	Flexible	420	88 (21.0)	119 (28.3)	213 (50.7)
I feel like I have to double check everything the hospital does to my baby	Consent	148	36 (24.3)	45 (30.4)	67 (45.3)
	Information	147	52 (35.4)	29 (19.7)	66 (44.9)
	Flexible	420	114 (27.2)	140 (33.3)	166 (39.5)
The hospital only provides screening tests that are important and safe	Consent	148	13 (8.8)	48 (32.4)	87 (58.8)
	Information	147	7 (4.8)	32 (21.8)	108 (73.4)
	Flexible	420	43 (10.2)	128 (30.5)	249 (59.3)
Most of medical researchers want to work on things that will make life better for the average person	Consent	148	11 (7.4)	43 (29.1)	94 (63.5)
	Information	147	7 (4.7)	21 (14.3)	119 (81.0)
	Flexible	420	34 (8.1)	115 (27.4)	271 (64.5)
If the government has funded a health test or procedure, it's probably a worthwhile test to have	Consent	148	14 (9.5)	49 (33.1)	85 (57.4)
	Information	147	9 (6.1)	44 (29.9)	94 (64.0)
	Flexible	420	39 (9.3)	131 (31.2)	250 (59.5)
The government wouldn't fund a health test or procedure if they were not sure of its benefits	Consent	148	23 (15.5)	46 (31.1)	79 (53.4)
	Information	147	19 (12.9)	35 (23.8)	93 (63.3)
	Flexible	420	49 (11.7)	143 (34.0)	228 (54.3)

Appendix Table 5.10: Cultural worldview, by class (Lachapelle et al., 2014), sub-scale scores, by class

Scale	Class	N	Mean	SD[1]	Min	Max
Hierarchism (2.74)	Consent	146	2.63	0.72	1	4
	Information	146	2.57	0.70		
	Flexible	420	2.84	0.66		
Individualism (2.53)	Consent	146	2.43	0.65	1	4
	Information	146	2.36	0.69		
	Flexible	420	2.62	0.70		
Egalitarianism (3.09)	Consent	146	3.18	0.76	1	4
	Information	146	3.19	0.78		
	Flexible	419	3.02	0.73		

Appendix Table 5.11: Participant demographics, by class

Demographics	Categories	N	Consent Priority	Information Priority	Flexible
Age N (%)	≤ 24 25 to 29 30 to 34 35 to 39 ≥ 40	$N_1 = 147$ $N_2 = 147$ $N_3 = 419$	4 (2.7) 27 (18.4) 54 (36.7) 35 (23.8) 27 (18.4)	6 (4.1) 16 (10.9) 44 (29.9) 49 (33.3) 32 (21.8)	26 (6.2) 68 (16.2) 139 (33.2) 110 (26.3) 76 (18.1)
Gender N (%)	Male Female Other	$N_1 = 147$ $N_2 = 147$ $N_3 = 419$	54 (36.7) 93 (63.3) 0 (0.0)	43 (29.2) 104 (70.8) 0 (0.0)	194 (46.3) 223 (53.2) 2 (0.5)
Indigenous status N (%)	Indigenous Not Indigenous Prefer not to answer	$N_1 = 147$ $N_2 = 146$ $N_3 = 418$	12 (8.1) 134 (91.2) 1 (0.7)	7 (4.8) 136 (93.1) 3 (2.1)	47 (11.2) 363 (86.8) 8 (1.9)
Ethnicity N (%)	White Chinese South Asian Black Filipino Latin American Southeast Asian Others	$N_1 = 146$ $N_2 = 147$ $N_3 = 419$	105 (71.9) 7 (4.8) 16 (11.0) 3 (2.0) 2 (1.4) 1 (0.7) 2 (1.4) 10 (6.8)	103 (70.1) 6 (4.1) 5 (3.4) 3 (2.0) 9 (6.2) 3 (2.0) 3 (2.0) 15 (10.2)	287 (68.5) 26 (6.2) 30 (7.2) 24 (5.7) 4 (0.9) 7 (1.7) 5 (1.2) 36 (8.6)
Marital status N (%)	Married or living as married Widowed Divorced Single-never married	$N_1 = 145$ $N_2 = 146$ $N_3 = 415$	128 (88.3) 0 (0.0) 4 (2.7) 13 (9.0)	139 (95.2) 2 (1.4) 0 (0.0) 5 (3.4)	364 (87.7) 5 (1.2) 9 (2.2) 37 (8.9)
Annual income N (%)	≤ 25,900 25,901 to 46,100 46,101 to 70,800 70,801 to 111,50 > 111,500	$N_1 = 147$ $N_2 = 146$ $N_3 = 420$	10 (6.8) 16 (10.9) 36 (24.5) 59 (40.1) 26 (17.7)	12 (8.2) 16 (11.0) 28 (19.2) 45 (30.8) 45 (30.8)	20 (4.7) 47 (11.2) 115 (27.4) 136 (32.4) 102 (24.3)
Highest education level N (%)	No post-secondary Trade certificate or diploma Community college University but no Bachelor's Degree Bachelor's Degree Post-graduate degree	$N_1 = 147$ $N_2 = 146$ $N_3 = 420$	18 (12.2) 15 (10.2) 18 (12.2) 16 (10.9) 46 (31.3) 34 (23.2)	20 (13.7) 14 (9.6) 29 (19.9) 10 (69.8) 49 (33.6) 24 (16.4)	41 (9.7) 42 (10.0) 57 (13.6) 73 (17.4) 134 (31.9) 73 (17.4)
Location N (%)	Rural Small city/town Medium and large city	$N_1 = 147$ $N_2 = 146$ $N_3 = 418$	16 (10.9) 35 (23.8) 96 (65.3)	24 (16.4) 27 (18.5) 95 (65.1)	55 (13.2) 69 (16.5) 294 (70.3)
Language spoken at home N (%)	English French Other	$N_1 = 146$ $N_2 = 145$ $N_3 = 418$	139 (95.2) 4 (2.7) 3 (2.1)	137 (94.5) 2 (1.4) 6 (4.1)	382 (91.4) 20 (4.8) 16 (3.8)
Number of children in household N (%)	One Two or more	$N_1 = 147$ $N_2 = 146$ $N_3 = 420$	76 (51.7) 71 (48.3)	60 (41.4) 85 (58.6)	219 (52.3) 200 (47.7)
Youngest child's place of birth N (%)	Hospital Birthing centre Home	$N_1 = 147$ $N_2 = 145$ $N_3 = 420$	135 (91.8) 9 (6.1) 3 (2.1)	142 (97.9) 2 (1.4) 1 (0.7)	369 (87.9) 38 (9.0) 13 (3.1)