A Public Health Ethics Analysis of Consent as a Least Restrictive Alternative for Newborn Screening in Ontario

by

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A thesis submitted in conformity with the requirements for the degree of Doctor of Philosophy

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Abstract

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Background: Public health ethics (PHE) is a theoretical lens used to analyze public health initiatives. PHE strives to find a balance between achieving population health goals and promoting individual liberty. To reach this balance PHE scholars encourage the use of least restrictive alternatives. However, least restrictive approaches are often taken for granted as presumed goods and their ability to protect individual liberty and the common good is often assumed. I examine this presumption through an examination of informed consent – implied and express – for newborn screening (NBS) in Ontario.

Methods: I conducted an exploratory qualitative case study to understand how 57 individuals involved in the lobbying, development, and implementation of expanded NBS in Ontario perceive consent policy for NBS. Semi-structured interviews were audio-recorded and transcribed. Data transformation was descriptive, analytic, interpretive, and applied.
Findings: Participants described their attitudes towards informed consent for NBS. PHE principles of least restrictive alternatives, effectiveness, autonomy, and social justice were key themes within implied consent data. Participants appreciated implied consent’s capacity to achieve high screening uptake, yet doubted its ability to generate informed decisions. Regarding express consent, participants introduced a host of concerns – interpreted as harm-causing – perceived to threaten NBS’s public health goals and individual autonomy. I applied the practice of consent to a PHE framework as a mechanism through which to consider consent policy for NBS in a way that moves towards achieving a balance between fulfilling public health goals and respecting individual rights and freedoms.

Conclusion: What participants in advisory capacities (and those to whom they turn for input) think about consent for NBS arguably influences their recommendations to the government. Challenging informed consent as a presumed good and submitting the practice of consent to the same ethical scrutiny as the NBS program itself legitimates the concerns of those wary of consent, illuminates the perceived benefits and risks of consent, and creates an opportunity to mitigate risks before implementing or adjusting consent policy. Treating the practice of consent as an intervention in theory could work to ensure that the ideals reflected in the concept of consent are realized in NBS practice.
Dedication

To my parents,
With love and gratitude
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Chapter 1. Introduction

*Introduction to Newborn Screening*

Universal newborn screening (NBS) has been in effect in North America since the early 1960s. Newborn screening was first used to screen infants for phenylketonuria (PKU), a rare inborn error of metabolism. Within a few years, screening for the endocrine disorder congenital hypothyroidism (CH) was added to the program (Tarini, 2007).¹ Left undiagnosed, these conditions can cause severe and irreversible physical and/or cognitive morbidity and/or mortality (Lloyd-Puryear & Forsman, 2002; Spady et al., 1998).

Identifying such disorders at birth facilitates medical management, enables early treatment, increases life-expectancy, reduces morbidity and mortality rates, and saves society in health care costs (Faden et al., 1982; Schoen et al., 2002). Over the years since it was first implemented, NBS has become one of the most widely supported preventive health interventions implemented throughout North America and Europe (American Academy of Pediatrics, 2000; Kwon & Farrell, 2000; Cunningham, 2002). In the 1990s, the advent of the tandem mass spectrometer² for NBS presented NBS programs with the possibility of

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¹ In Ontario, infants were screened for PKU in 1965 and 1978 for CH (Newborn Screening Ontario, n.d.a).

² The tandem mass spectrometer is an analytical instrument used to identify compounds (amino acids and acylcarnitines) found in an infant’s blood. If amino acids and acylcarnitines are not metabolized properly, these compounds will amass in the baby’s blood and tissues and ultimately put the infants’ health and/or life at risk. Tandem mass spectrometry measures multiple compounds simultaneously to determine whether too much of one compound exists in any given infant (Natowicz, 2005; Waisbren, 2003). This technology has the capacity to screen more than 400 samples per day and is extremely accurate, offering 100% sensitivity and a specificity rating of 83% to 99% depending on the condition being screened (Hanley, 2005; Medical Advisory Secretariat, 2003). The technology promises reduced false positive rates, improved cost-benefit ratio and overall superior screening outcomes in contrast to earlier NBS methods (Hanley, 2005; Medical Advisory Secretariat, 2003). Schoen et al. (2002) argue, however, that even though expanded NBS is comparable to other screening and treatment programs, tandem mass spectrometry yields higher false-positive results given the added number of conditions for which newborns are screened.
expanding significantly the number of conditions for which infants could be screened (Schoen et al., 2002). This technology, however, generated significant public debate concerning perceived ethical issues related to the expansion of NBS programs (Dhanda & Reilly, 2003; Hanley, 2005; Guthrie, 2005). Specifically, questions were raised as to how many conditions should be added to the screening panel given the strengths and limitations of the technology and the limited treatment options for many of the conditions that could be detected through screening (Wilken, 2003; Natowicz, 2005). Other concerns pertained to the increased number of false positives that would be generated as a result of the technology (Schoen et al., 2002; Medical Advisory Secretariat, 2003).

Given the cost of acquiring the new technology and the uncertainty associated with its implementation, not all states, provinces or countries with NBS programs chose to expand (Dhanda & Reilly, 2003). However, as new conditions were able to be diagnosed, advocacy groups demanded national uniform screening programs (Guthrie, 2005). Variability in newborn screening practices challenged public health departments to address issues of justice, fairness, and equitable distribution of resources (Guthrie, 2005; Eggertson, 2005). Regional variations in screening programs led to calls for standardization of screening initiatives (Stoddard & Farrell, 1997; Baily, 2009).

Gradually, governments internationally have articulated their commitment to promote the health of individuals with rare conditions by funding expanded NBS programs. Today, many programs have evolved to incorporate technology that can screen newborns for upwards of 30 conditions, identify carrier status, aid in reproductive decision-making, store dried newborn blood spots, and facilitate research. Despite technological and conceptual
changes in NBS, the approach to parental consent in North America has largely remained unchanged.

**Debate Around Consent for Newborn Screening**

In the United States the vast majority of states have legislation mandating NBS and in Canada NBS programs operate according to an implied consent approach whereby parental consent is inferred.\(^3\) Mandatory\(^4\) and implied consent approaches to NBS have been heavily criticized since the inception of NBS in the 1960s and the debates have only intensified with program expansion (Annas, 1982; Paul, 1997; Wildeman & Downie, 2001; Ross, 2010). Now, with the increase in the number of conditions for which infants are screened, questions around the effectiveness and long-term benefit of treatment options for some of the conditions on the screening panel, an increased number of incidental findings, the ability to identify carriers of sickle cell anaemia trait, the long-term storage of dried blood spots and future uses for such blood spots (e.g., research, forensics), some scholars argue that the benefit-to-harm ratio of NBS has shifted, further challenging mandatory and implied consent

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\(^3\) See Appendix A for a sample of jurisdictions and their respective approaches to consent for NBS.

\(^4\) “Mandatory” or “compulsory” are labels given to those screening programs that have enacted legislation mandating that infants be screened (Wildeman & Downie, 2001). (The language of “mandate” in public health, however, is not limited to legislated measures; rather, “mandate” is often used rhetorically either to challenge public health interventions that have become routine or to underscore the importance of the intervention and/or to encourage insurers to assume the costs (Wynia, 2007).) In jurisdictions that have mandatory screening, particularly in the United States, many allow parents to opt-out – or decline screening – for religious, personal, or philosophical reasons (Therrell *et al.*, 2006; Newson, 2006). However, a common criticism of such an approach is that very often parents do not know that their infants are screened and, consequently, do not know that they can opt-out (Hiller *et al.*, 1997; American Academy of Pediatrics, 2000; Committee on Bioethics, 2001 Campbell & Ross, 2003; Wilcken, 2003; Dhanda & Reilly, 2003). The nature of mandatory public health programs has shifted over the decades. There was a time when parents who failed to comply with state-enforced mandates would be fined or incarcerated (Gostin, 2005). Today, most “mandatory” public health interventions allow individuals to opt-out—although the difficulty with which one can opt-out varies from a simple “no” to more extreme bureaucratic hurdles (Wynia, 2007: 3). Although, as Wynia (2007) asks, if individuals can opt-out of a public health intervention for any reason, to what extent is it mandatory?: “is something mandatory if you don’t really have to do it?” (p. 3). For the purposes of this study, I will use the language of “mandatory screening” in reference to those jurisdictions that have legislation mandating – with or without opt-out clauses – NBS thereby requiring parents to have their infants screened.
approaches for NBS (American Academy of Pediatrics, 2000; McCabe et al., 2002; Liebl et al., 2002; Wilcken, 2003; Waisbren et al., 2003; Campbell & Ross, 2003; Natowicz, 2005; Botkin, 2005). The amount of information generated as a result of NBS technology coupled with emergent issues related to storage and research heightens concerns around issues of privacy and confidentiality as well as concerns around genetic discrimination and stigmatization by insurance companies, employers, hospitals, and even an individual’s community and/or family members (American Academy of Pediatrics, 2000; McCabe et al., 2002; Mandl et al., 2002; Dhanda & Reilly, 2003; Leberge et al., 2004). As newborn screening technology continues to advance, so too do the possibilities for how the technology can be used within the program and its subsequent effects on individuals, families, and the community at large. These developments strengthen arguments against mandatory and implied consent approaches in favour of a more explicit approach to parental consent.

Among the main criticisms of mandatory screening and implied consent for NBS is that many parents across different jurisdictions remain uninformed or underinformed about NBS and, as a result, are often unaware that they have the ability to opt-out (Hiller et al., 1997; American Academy of Pediatrics, 2000; Committee on Bioethics, 2001; Wildeman & Downie, 2001; Campbell & Ross, 2003; Wilcken, 2003; Dhanda & Reilly, 2003; Ross, 2010; Araia et al., 2012). In Ontario, Canada, NBS occurs through implied consent (Miller et al., 2010) which, according to the Health Care Consent Act 1996, is considered an acceptable approach to informed consent: “Consent to treatment may be express or implied” (c.2, Sched. A, s. 11 (4)). However, consent “must be informed”: patients or surrogate decision makers must be educated about the type of treatment, the benefits and risks of treatment, the
alternatives to treatment, and the potential implications of not having the treatment (*Health Care Consent Act 1996*, c. 2, Sched. A, s. 11(3)). Unlike express consent which “is directly given, either orally or in writing,” implied consent “occurs when surrounding circumstances are such that a reasonable person believes that consent has been given, although no direct, express or explicit words of agreement had been uttered” (College of Physicians and Surgeons of Ontario, 2006: 5). Important to underscore, however, is that the test is not whether the patient (or any reasonable patient) would have consented on a reasonable analysis of these or other circumstances, but rather whether it was reasonable for the health care provider to infer from the circumstances that the patient did in fact consent. (Wildeman & Downie, 2001: 94-95).

Therefore, challenges to parental awareness, knowledge, and understanding of NBS in jurisdictions such as Ontario that use an implied consent approach have significant implications for whether HCPs can “infer consent from an absence of refusal” (Wildeman & Downie, 2001: 100). In fact, the difficulty facing HCPs to provide “[e]vidence of [c]onsent” with implied consent led the College of Physicians and Surgeons of Ontario (2006) to discourage implied consent in favour of express consent: “Although the Act states that consent to treatment may be express or implied, physicians are strongly advised to obtain express consent from the patient” (p. 5).

Although NBS programs in North America remain predominantly rooted in mandatory or implied consent screening traditions, countries in Europe, Asia, and Australasia are making efforts to implement more express consent practices for NBS in their respective jurisdictions (see Appendix A for a sample of jurisdictions and their respective approaches to consent for NBS). Given that in Ontario informed consent may be achieved either through implied consent or express consent (*Health Care Consent Act, 1996*; College of Physicians
and Surgeons of Ontario, 2006), for clarity purposes in this thesis I will be using the language of express consent, defined as consent that is “directly given, either orally or in writing. It is positive, direct, unequivocal consent, requiring no interference or implication to supply its meaning” (College of Physicians and Surgeons of Ontario, 1996: 5). However, “informed consent” or “informed choice” are the two most frequently used characterizations of NBS programs that require explicit parental consent in contrast to implied consent. In the NBS literature, these informed consent labels and definitions differ (Hargreaves et al., 2005b); yet, commonalities across the definitions reflect the historical roots of the concept – originally called “voluntary consent” – which date back to 1947 with the Nuremberg Code and its Directives for Human Experimentation:

The voluntary consent of the human subject is absolutely essential. This means that the person involved should have legal capacity to give consent; should be so situated as to be able to exercise free power of choice, without the element of force, fraud, deceit, duress, over-reaching, or other ulterior form of constraint or coercion; and should have sufficient knowledge and comprehension of the elements of the subject matter involved as to enable him to make an understanding and enlightened decision. This latter element requires that before the acceptance of an affirmative decision by the experimental subject there should be made known to him the nature, duration, and the purpose of the experiment; the method and means by which it is to be conducted; all inconveniences and hazards reasonable to be expected; and the effects upon his health or person which may possibly come from his participation in the experimentation. (p.181)

The requirements for voluntary consent outlined in the Nuremberg Code (1947) for research contexts extend into the clinical realm (Cassileth et al., 1980; Faden & Beauchamp, 1986) and in public health contexts as well, as evidenced by how scholars have engaged with the definition of informed consent for NBS (Hargreaves et al., 2005b; Nicholls, 2010). An

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5 Although I use the label of express consent to contrast implied consent in my dissertation, during the interviews my participants and I used the informed consent label as the direct contrast to implied consent.

6 Ross (2010) used the phrase “voluntary consent.”
important emphasis in the NBS literature addressing the issue of informed consent is the
focus on the necessary requirement for patient or surrogate decision-maker understanding:

Such choice or consent is informed if the person ‘receives a thorough disclosure about the
procedure, comprehends the disclosed information, acts voluntarily, is competent to act,
and consents.’ (Beauchamp and Faden, 1995, p 1239). A competent person may, therefore, action a choice, but this may not be informed if the individual does not
‘comprehend the disclosed information.’ As a result understanding is central to the
‘informed’ element of an informed choice. (Nicholls, 2010: 129)

Decisions are made voluntarily; By parents who have considered and understood
information about the screening, including the benefits and risks. (Hargreaves et al.,
2005b: 162)

The basis of informed consent is that the patient receives and understands enough
information about the testing process and the condition being tested for, to make an
informed decision about whether to undergo testing. (Provincial Advisory Committee on
New Predictive Genetic Technologies, 2001: 35)

These definitions of informed consent within the context of NBS reiterate the importance of
parental knowledge and understanding as a critical requirement to ensure, to the degree
possible, valid informed consent. However, as with criticisms directed towards mandatory
and implied consent screening programs, some scholars engaged in NBS research in
jurisdictions that purport to obtain express parental consent have reported a lack of parental
awareness, knowledge, and/or understanding about NBS as well (Huang et al., 2005;
Hargreaves et al., 2005b; Davis et al., 2006). Such challenges to the practice of express
consent in terms of it being able to succeed in achieving informed, autonomous decisions are
mirrored in consent research in both clinical and research contexts (Flory & Emanuel, 2004;
Tait et al., 2005; Dawson, 2005; Crepeau et al., 2011).

Despite the challenges of express consent in practice, the technological and
procedural expansion of NBS – particularly as it pertains to the storage of and research on
dried newborn blood spots – arguably forces jurisdictions that operate NBS programs using mandatory or implied consent mechanisms to reevaluate their approach to parental consent for NBS (Ross, 2010). Additional arguments in support of express parental consent for NBS are often rooted in legal, philosophical, and moral arguments underscoring individuals’ right to self-determination with respect to health care decisions and parents’ authority as the most appropriate surrogate decision-makers for their infants (Wildeman & Downie, 2001; Committee on Bioethics, 2001; Huang et al., 2005; Ross, 2010).7

In Ontario, formal and informal advisory committees for NBS have been in existence over the years to offer recommendations to the government on issues pertaining to the NBS program’s policies, guidelines, and practices. Scholars have explicitly called for the Ontario government to examine the province’s consent policy for NBS since 2001. Ontario’s

7 In Canada, the common law and certain provincial statutes grant parent(s) or guardian(s) the legal “[a]uthority to consent to treatment of an infant or minor who is incapable of making a treatment decision” (Wildeman & Downie, 2001: 89):

“[t]he common law has long recognized that parents are in the best position to take care of their children and make all the decisions necessary to ensure their well-being….This recognition was based on the presumption that parents act in the best interest of their child’ (Supreme Court of Canada decision B.(R) v. Children’s Aid Society of Metropolitan Toronto [1995] I S.C.R. 315 at 370, cited in Wildeman & Downie, 2001: 89).

This authority, however, is not without limits; parents are required to make “health care decisions that are in the best interests of their child” (Wildeman & Downie, 2001: 90):

Parents who fail to provide their children with necessary medical treatment may be subject to criminal liability as well as child protection proceedings. In addition, an application may be made to the court to exercise its parens patriae jurisdiction to protect the child’s best interests (Wildeman & Downie, 2001: 90).

The state is bound by limitations as well, permitted only to intervene in “the most extreme cases”:

“In recent years, courts have expressed some reluctance to interfere with parental rights, and state intervention has been tolerated only when necessity was demonstrated. This only serves to confirm that the parental interest in bringing up, nurturing and caring for a child, including medical care and moral upbringing, is an individual interest of fundamental importance to our society’ (Supreme Court of Canada decision B.(R) v. Children’s Aid Society of Metropolitan Toronto [1995] supra note at 370-371 cited in Wildeman & Downie, 2001: 90-91). Although the courts may, in the end, overrule parental decisions pertaining to their children’s health care deemed “unreasonable”, parents are legally “allowed, at least in the first instance, to make unreasonable decisions about the medical care of their children. However, there are mechanisms for challenging those decisions in court” (Wildeman & Downie, 2001: 107):

The reasonableness argument may well have some legal force in a courtroom — but it is in the courtroom, and not in the hands of health care professionals as such, that the force of such an argument must be determined. Consent must be sought despite the fact that it may ultimately be overridden by an exercise of the court’s parens patriae jurisdiction. (Wildeman & Downie, 2001:107)
Provincial Advisory Committee on New Predictive Genetic Technologies wrote in their 2001 report, *Genetic Services in Ontario: Mapping the Future*, that future committees examining NBS in Ontario needed to pay particular attention to the issue of consent. This committee explicated the need for express, written, parental consent:

> As a general rule, parental consent should be required for newborn genetic screening. The Provincial Advisory Committee, or another body accountable to the government, should investigate whether, and under what circumstances, newborn screening without express parental consent is permissible. (Provincial Advisory Committee on New Predictive Technologies, 2001: 74)

Similarly, Wildeman and Downie (2001) examined whether NBS programs that operate without obtaining explicit parental consent are “legally defensible” and ultimately concluded that either practice or legislation needs to change:

> In the end, we conclude that either practice should be changed to align it with current law such that explicit parental consent is sought for the established tests, or that advocates for maintaining current practices should lobby for legislation permitting newborn screening in the absence of explicit parental consent. The approach to the issue of consent to the new tests can then be built upon a legally defensible foundation. (p. 63)

As of Spring 2010, the policy and practice of consent for NBS was officially put on the Maternal-Child Screening Committee’s agenda — the province’s current advisory committee (Born Ontario, 2013a).

A growing body of Ontario-based NBS scholarship has already begun to contribute to this debate. Past studies have focused on consent for carrier status disclosure (Hayeems *et al.*, 2008; Miller *et al.*, 2009a; Miller *et al.*, 2009b; Miller *et al.*, 2010a); perceptions of consent for NBS from the perspectives of parents and health care providers (HCPs) (Bombard *et al.*, 2012; Miller *et al.*, 2010b); HCPs’ perceptions of their capacity to inform
parents about NBS (Hayeems et al., 2009); and perceptions of NBS education from the perspectives of parents (Araia et al., 2012).

**Study Goals and Objectives**

My qualitative case study aims to contribute to this discussion through an examination of attitudes and perceptions towards informed consent – both implied and express – from the perspectives of key informants engaged in Ontario’s NBS program and responsible for advising the government, either directly or indirectly, on the policy and practice of consent for NBS in Ontario. Drawing on public health ethics principles of least restrictive alternatives, autonomy, effectiveness, and social justice I offer a public health ethics analysis of the phenomenon of consent for NBS with a particular focus on the tensions between respecting parental autonomy and achieving the public health goals of NBS. This inquiry challenges the perceived effectiveness of informed consent – both implied and express – as a least restrictive alternative for NBS and offers a public health ethics approach to examine consent policy and practice for NBS in Ontario, Canada. In short, this dissertation explores:

1. Participants’ perceptions of the goals and objectives of NBS in Ontario;
2. Participant attitudes and perceptions towards the current implied consent approach to NBS in Ontario;
3. Participant attitudes and perceptions towards express consent for NBS, including the storage and research dimensions of this expanded program;
4. Participant ambivalence towards express consent for NBS; and
5. The usefulness of a public health ethics lens to examine consent as a least restrictive alternative for a public health intervention such as NBS.
Before I outline what to expect in each of the subsequent chapters in this thesis, I will provide some background about Ontario’s NBS program.

**Newborn Screening in Ontario: Background**

Until April 2006, Ontario’s NBS program screened only for phenylketonuria (PKU) and congenital hypothyroidism (CH). After years of lobbying, the Ministry of Health and Long-Term Care announced on November 2, 2005 that Ontario’s provincial NBS program would be expanding (Ministry of Health and Long-Term Care, 2005a). The province implemented an incremental expansion. By April 3, 2006 infants were screened for PKU, CH, and MCAD (Medium Chain Acyl-Co-A dehydrogenase) deficiency (Newborn Screening Ontario, n.d.a). Eighteen additional metabolic conditions were added to Ontario’s newborn screen on August 8, 2006 (Newborn Screening Ontario, n.d.a). On November 24, 2006 screening commenced for three hemoglobin disorders: Sickle Cell Anemia (HbSS), Sickle Hemoglobin C Disease (HbSC), and Sickle Beta Thalassemia (HbS/ß-Thalassemia) (Newborn Screening Ontario, n.d.a). Biotinidase deficiency and galactosemia were added to the screening panel on February 28, 2007 and congenital adrenal hyperplasia was added on May 14, 2007 (Newborn Screening Ontario, n.d.a). Cystic fibrosis was added to this list on April 7, 2008, bringing the number of conditions for which Ontario-born infants are screened to twenty-eight (Newborn Screening Ontario, n.d.a). As of November 1, 2010, parents can now actively request to receive their infant’s carrier status results for sickle cell anaemia “and some less common hemoglobinopathies” (Newborn Screening Ontario, n.d.a). Today in Ontario, approximately 140,000 infants are screened annually for 28 rare metabolic, endocrine, and hemoglobin conditions through Newborn Screening Ontario, the province’s
NBS program. Approximately 150 out of 140,000 infants born each year in Ontario will be affected by one of the conditions on the NBS panel (Newborn Screening Ontario, n.d.b).

The Screening Process

The screening process typically occurs between 24 hours and 7 days after birth (Newborn Screening Ontario, n.d.c). Infants’ heels are pricked and several drops of blood are collected on filter paper (or blood spot/specimen cards). The filter paper is then mailed to the Newborn Screening Ontario Laboratory at the Children’s Hospital of Eastern Ontario in Ottawa for analysis. It typically takes two weeks to receive the NBS results (Newborn Screening Ontario, n.d.k). The parents of infants who screen positive for one of the conditions on the screening panel are contacted immediately and follow-up testing is scheduled, usually through one of six regional NBS treatment centres for follow-up diagnostic testing and, if necessary, treatment (Newborn Screening Ontario, n.d.c).

After screening the dried blood spots are stored in Ontario for 19 years as part of each child’s medical record, after which point the samples are destroyed (Newborn Screening Ontario, n.d.f). The primary reason for storing the dried blood spots in Ontario is for quality control and quality assurance to maintain and improve the NBS system (Newborn Screening Ontario, n.d.f). Other reasons for blood spot storage include being able to retest the sample in the event the infant gets sick; use by the Coroner’s office in the event of unexpected infant death; sending part of the sample to another laboratory for further testing upon the written

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8 Later this year a 29th condition – severe combined immunodeficiency (SCID) – will be added to Ontario’s NBS panel (Newborn Screening Ontario, n.d.h).

9 There are five regional treatment centres in Ontario (Children’s Hospital of Western Ontario, McMaster Children’s Hospital, The Hospital for Sick Children, Kingston General Hospital, and Children’s Hospital of Eastern Ontario) and one in Winnipeg, Manitoba for those infants who screen positive in North-Western Ontario (Newborn Screening Ontario, n.d.d).
request of a parent or guardian; and conducting de-identified research (Newborn Screening Ontario, n.d.f). Parents may request for the immediate destruction of their infants’ dried blood spots (Newborn Screening Ontario, n.d.f). However, at the recommendation of the 2008 task force of the Ontario Advisory Committee on Newborn and Childhood Screening, parents are advised to keep their infants’ blood spots in storage for at least 5 years since the conditions for which infants are screened typically manifest by 5 years of age (Newborn Screening Ontario, n.d.f). A stored blood spot “would allow investigation and possible re-testing if a child was diagnosed with one of the conditions on our [Newborn Screening Ontario’s] panel following a negative screen” (Newborn Screening Ontario, n.d.f).

**The Public Health Roots of Newborn Screening in Ontario**

The changes to Ontario’s NBS program in 2006 were not limited to the implementation of the tandem mass spectrometer and the increased number of conditions for which infants born in the province are screened. Rather, the physical location of the program shifted from its home of 40 years in the Public Health Branch of the Ministry of Health and Long-Term Care (now Public Health Ontario) to the Children’s Hospital of Eastern Ontario in Ottawa. The history of NBS as a public health program entrenched in public health principles (Faden *et al.*, 1982; Beauchamp & Steinbock, 1999; Baily & Murray, 2009), led a number of former Advisory Committee members to express their dissatisfaction with the Public Health Branch’s perceived refusal to recognize NBS as a public health issue:

On December 14, 2004, a number of the former Advisory Committee members wrote to Dr. Basrur, Chief Medical Officer of Health stating:

We have been and continue to be disappointed at the refusal of the PHD (Public Health Division) to acknowledge that universal newborn screening for the people of Ontario is
most definitely a public health issue and is seen as such by most other jurisdictions worldwide. (Marin, 2005: 21)

Newborn Screening Ontario has since articulated that NBS in Ontario remains rooted in a public health philosophy:

Newborn screening: The big picture Newborn screening in Ontario has seen a dramatic overhaul in the past few years. In 2005, the program shifted from a Public Health Laboratory to a dedicated Newborn Screening Program based at CHEO in Ottawa. ‘The Program currently screens every baby born in Ontario for 27 diseases. Expanding the program from its original panel of two diseases involved a tremendous effort and commitment from many people at CHEO and across the province. Similar expansions are occurring in provinces across Canada and we’re hopeful that they will be adding CF to their programs as well,’ notes Dr. Chakraborty.

The philosophy remains unchanged; screening attempts to identify treatable disorders in neonates that, if undetected, would lead to mental retardation or have life-threatening consequences. The benefits of early detection and treatment will soon extend to CF, improving the prognosis of children born with this complex disease. (Honeywell & Cloutier, 2007)

The commitment to NBS’s public health origins were arguably reinforced when Newborn Screening Ontario — under the stewardship of the Ministry of Health and Long-Term Care — continued its implied consent policy for NBS for the expanded program:

Historically, newborn screening (NBS) has been a routinized intervention that is either legislated (e.g., most US states) or operates with implied consent (e.g., Canada). This approach has its origins in the legacy of infant screening as a mandated ‘public health emergency’, with the severity and treatability of exemplar conditions like phenylketonuria (PKU) and congenital hypothyroidism (CH) obliging early detection. (Miller et al., 2010b: 181)

Implied consent for NBS in Ontario includes the newborn screen, storage of dried blood spots, and use of dried blood spots for de-identified research (Bombard et al., 2012; Newborn Screening Ontario, n.d.f).
The “police powers” granted public health officers in certain health situations to promote the health and safety of the public\textsuperscript{10} have arguably been preserved in NBS by the Children’s Aid Society. Specifically, on Newborn Screening Ontario’s website, parents are alerted to the fact that while NBS is not mandatory and that they may decline screening on behalf of their infant, the Children’s Aid Society may be called should they choose to do so:\textsuperscript{11}

\textbf{As a parent, may I refuse to have the NBS test done?}

Newborn screening is designed to identify serious diseases in infants at an early age, so that early treatment can be provided to provide the best outcomes for babies with these diseases. Newborn screening is considered standard of care for your baby and is highly recommended; any decision to decline testing should first be discussed with a health care provider. Newborn screening is not mandatory in Ontario. The hospital where you give birth or your health care provider may ask you to sign a form indicating that you have declined the NBS and may notify Newborn Screening Ontario (NSO). Some hospitals or health care providers have indicated that they may opt to contact the Children's Aid Society if parents decline newborn screening.

The benefits of newborn screening for babies far outweigh any of the theoretical risks associated with having the blood sample stored. About 200 babies are born with one of the 28 diseases screened for every year in Ontario. Most of the parents of these babies had no idea their baby could have one of these diseases. These babies have the chance to live longer, healthier lives because of newborn screening. (Newborn Screening Ontario, n.d.g)\textsuperscript{12}

\textsuperscript{10} For more on public health’s “police powers” please turn to Chapter Two.

\textsuperscript{11} One might reasonably question the voluntariness of a program that threatens a visit from the Children’s Aid Society if parents decline NBS.

\textsuperscript{12} This excerpt was accessed from the website on October 18, 2012. Since I began my inquiry the response to this question found under the Frequently Asked Question section of the website (Newborn Screening Ontario, n.d.g) has evolved. On August 13, 2009 the response read:

\textbf{As a parent, may I refuse to have the NBS test done?}

Newborn screening is designed to identify serious diseases in infants at an early age, so that early treatment can be provided to provide the best outcomes for babies with these diseases. Newborn screening is considered standard of care for your baby and is highly recommended; any decision to decline testing should first be discussed with a health care provider. The hospital where you give birth or your health care provider may ask you to sign a form indicating that you have declined the NBS and may notify the Ontario Newborn Screening Program. Some hospitals or health care providers have indicated that they may opt to contact the Children's Aid Society if you refuse NBS for your baby.

The underlying message, however, has remained consistent: a decision to decline screening on behalf of one’s infant may involve reporting the decision to the Children’s Aid Society.
In addition, the Children’s Aid Society, the Royal Canadian Mounted Police, and Public
Health Ontario may be called to help track down infants who have screened positive for one
of the conditions on the newborn screen in instances where the treatment centres are unable
to locate the families (Newborn Screening Ontario Clinical Follow-Up Symposium, 2010).

Given Ontario’s history of NBS as a public health program, Newborn Screening
Ontario’s sustained commitment to honouring the public health principles of NBS, and the
continued practice of implied consent for NBS in the province, a public health ethics inquiry
of consent for NBS in Ontario will offer a unique perspective into the exploration of key
informant attitudes and perspectives towards the policy and practice of consent for NBS in
Ontario.

Organization of the Thesis

This dissertation consists of nine chapters. In Chapter Two, I provide an overview of
public health ethics, the conceptual framework that informed my inquiry from the outset.
Here I explain what public health ethics is and examine its usefulness for exploring public
health programs, interventions, and practices. I identify and explain four public health ethics
principles that recur throughout the thesis: least restrictive alternatives, effectiveness,
autonomy, and social justice. The final segment of this chapter explains my rationale for
applying a modified version of Public Health Ontario’s (2012) public health ethics
framework entitled, *A Framework for the Ethical Conduct of Public Health Initiatives*, to my
findings on participant attitudes and perceptions of consent for NBS.

Chapter Three provides an overview of the extant literature on consent for NBS.
Included in this review is empirical scholarship that explores the issue of consent for NBS
from the perspectives of parents and/or HCPs charged with (or would be charged with) obtaining such consent. These perspectives serve to complement and challenge my study findings. In addition, while much of the existing NBS scholarship has not used a public health ethics lens, I demonstrate that the four principles of public health ethics identified in my data are evident in these findings as well. Given that NBS programs have traditionally been considered public health programs, it is not surprising that one can identify principles of public health ethics in other studies. The presence of these principles in existing scholarship reinforced the appropriateness of a public health ethics inquiry of the policy and practice of consent for NBS.

Chapter Four offers a comprehensive overview of my methodology. I provide a detailed explanation of my study design, recruitment strategies, data collection, data analysis and interpretation, rigour, and ethics.

In Chapters Five through Seven I present the qualitative findings of my study. In Chapter Five I provide an overview of my participant demographics. The participants who volunteered to participate in my study fit largely into four categories: past and present members of NBS advisory committees; HCPs at one of the 5 NBS treatment centres across the province; parent-, patient-, HCP-, and community-advocates involved in NBS in some capacity; and legal experts identified by participants as being perceived to have contributed directly or indirectly to the debate regarding consent for NBS in Ontario.

In Chapter Six I present participants’ perceptions of implied consent for NBS: the current policy for NBS in Ontario. This chapter addresses the perceived strengths and weaknesses of an implied consent policy for NBS in Ontario as currently practiced.
In Chapter Seven I focus on participants’ attitudes and perceptions towards the possibility of introducing an express consent approach for NBS in Ontario. In this chapter I explore participants’ attitudes of express consent for NBS in relation to the perceived goals and objectives of the NBS program, storage of dried blood spots, and the use of these dried blood spots for research.

In Chapter Eight, I apply a modified version of Public Health Ontario’s (2012) public health ethics framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, to my findings on participant perceptions of express consent for NBS. This analysis has three goals: 1) to challenge some of the taken-for-granted assumptions embedded within the practice of consent within the context of NBS; 2) to highlight micro and macro issues to consider regarding the implementation of the practice of consent for NBS in Ontario; and 3) to provide a public health ethics framework that could be useful to the advisory committees as they continue to debate issues of consent pertaining to storage of and future research on dried newborn blood spots.

In the final chapter of my dissertation, Chapter Nine, I present what I perceive to be the most salient contributions of my inquiry to the ongoing debate about consent for NBS and public health ethics scholarship. Based on my findings I also offer recommendations to the advisory committees to consider the inclusion of professionals with different expertise to complement the existing composition of the committee as well as ideas for future research in this area.
Chapter 2. Conceptual Framework

Chapter Overview

Public health ethics is the lens through which I designed my study and analyzed and interpreted my data. I divided this chapter into two parts to explain what public health ethics is and why and how I used this perspective for my inquiry. Part I serves as an introduction to public health ethics. In this section I discuss what public health ethics is and why it is a useful lens to examine public health programs, policies, and interventions. I also describe the different ways in which scholars have contributed to this evolving field over the years. Finally, I end this section by examining the role and function of public health ethics frameworks. I explicate the goals and objectives of public health ethics frameworks and explain why they can be useful analytic tools for ethical analyses of public health initiatives.

Part II of this chapter explains how I applied this public health ethics lens to my research. First, I articulate my rationale for using a public health ethics lens to explore stakeholder perceptions of ethical issues related to expanded NBS in Ontario and their attitudes towards the policy and practice of consent for NBS in particular. Second, I explain and justify the two-pronged approach I used for my public health ethics inquiry: the identification of the public health ethics principles embedded in participant perceptions of and attitudes towards the policy and practice of consent for NBS followed by an application of these public health ethics principles of consent to an analytic public health ethics framework.
**Part I: An Introduction to Public Health Ethics**

*What is Public Health Ethics?*

Public health ethics is informed by moral and political philosophy and offers approaches to moral decision-making for public health programs, policies, and interventions (Holland, 2007; Baum *et al.*, 2007; ten Have *et al.*, 2010; Lee, 2012; ten Have *et al.*, 2013). Public health’s commitment to promoting social welfare can risk sacrificing the rights and freedoms of individuals for the greater good of the population, thereby generating unique ethical dilemmas unsuited to more traditional clinical ethics assessments (Beauchamp, 1999; Horner, 2000; Callahan & Jennings, 2002; Levin & Fleishman, 2002; Childress *et al.*, 2002; Jennings, 2003; Krantz *et al.*, 2004; Dawson & Verweij, 2007; Holland, 2007; Baum *et al.*, 2007; Swain *et al.*, 2008; Baylis *et al.*, 2008; ten Have *et al.*, 2010; Lee, 2012; ten Have *et al.*, 2013). The priority placed on the individual coupled with a predominant focus on biomedicine, biotechnology, and clinical care were among the rationales behind academic appeals for ethical analyses sensitive to the broader social mandate of public health and the inherent competing interests at play between individuals and the communities within which they live (Beauchamp, 1999; Childress *et al.*, 2002; Callahan & Jennings, 2002; Upshur, 2002; O’Neill, 2002; Benatar, 2003; Mackie & Sim, 2004; Nixon, 2006a; Dawson & Verweij, 2007; Holland, 2007; Baum *et al.*, 2007; Baylis *et al.*, 2008; Dawson, 2010; Dawson, 2011).

The diversity in approaches to moral theory generally, and bioethics more specifically, persists in the developing field of public health ethics (Sherwin, 1999; Beauchamp & Steinbock, 1999; Childress *et al.*, 2002; Callahan & Jennings, 2002; Upshur, 2002; Holland, 2007; Baylis *et al.*, 2008). Public health ethics has been influenced by John Stuart Mill and his articulation of the harm principle (Upshur, 2002; Childress *et al.*, 2002;
Nuffield Council on Bioethics, 2007; Holland, 2007); principlism (Upshur, 2002; Childress et al., 2002; Thompson et al., 2006; Miller et al., 2009a); virtue ethics (Horner, 2000; Holland, 2007); and human rights scholarship (Mann et al., 1999; Nixon & Forman, 2008; Benatar, 2003) to name a few. The undeniable social component of public health has, however, generated agreement among academics that public health ethics necessarily requires approaches to analyses that integrate community-oriented values (Callahan & Jennings, 2002; Roberts & Reich, 2002; Powers & Faden, 2006; Gostin & Powers, 2006; Dawson & Verweij, 2007; Thompson et al., 2003; Baylis et al., 2008; Kenny et al., 2010). Although public health is influenced to a significant degree by consequentialism, individual rights are important as well (Holland, 2007; Nixon & Forman, 2008; Jaffe & Hope, 2010):

[T]here is a consensus that the distinctive core of public health ethics is the trade-off that can arise between, on the one hand, protecting and promoting the health of populations, and, on the other, avoiding individual costs of various kinds, including physical danger, moral harm and frustrated desires. (Holland, 2007: ix)

Negotiating a balance between the individual and the community is a persistent challenge for public health policy making (Callahan & Jennings, 2002; Childress et al., 2002; Krantz et al., 2004; Gostin & Powers, 2006; Dawson & Verweij, 2007; Holland, 2007; Lee, 2012).

Finding “The Balance” in Public Health Ethics

Scholars argue that pursuing public health goals at the expense of individual rights and freedoms or protecting individual rights and freedoms without considering broader population health benefits and/or risks are “two equally unsatisfactory extremes” (Horner, 2000: 50; see also Holland, 2007). Policy makers (and their advisors) charged with decision-making around public health programs, policies, and interventions must grapple with these tensions in an effort to find a balance between these two extremes:
The Doctrine of the Mean in public health involves sensitively balancing the claims of individuals and the claims of communities rather than rushing to adopt policies that can meet only one at the expense of the other. (Holland, 2007: 34)

Holland (2007) cautions against allowing ethical discussions and debates to revert to “mere re-description of the central dilemma between individuals and communities” (p.189) — specifically, simply privileging the rights of one over the other — rather than working towards a resolution.

Part of working towards achieving this balance requires establishing the extent to which state interference is necessary to achieve a given public health goal (Upshur, 2002; Childress et al., 2002; Thompson et al., 2006; Baylis et al., 2008). Public health as a profession is bestowed with what are often referred to as “police powers,” defined as legislative authority bestowed on the state to issue coercive mandates that infringe on individual liberties in an effort to protect the welfare of the population (Callahan & Jennings, 2002; Childress et al., 2002; Upshur, 2002; Jennings, 2003; Thompson et al., 2006; Gostin & Powers, 2006; Holland, 2007; Baylis et al., 2008):

Public health is one of the few professions that has, in many matters, legal power— in particular, the police power of the state— behind it. It can, through the use of the law, coerce citizens into behaving in some approved healthy way […] Public health also has the distinction, along with a few others—such as city management, public administration, and law enforcement—of being a profession in which many practitioners are government employees and officials. It thus has an obligation both toward government, which controls it, and toward the public that it serves. (Callahan & Jennings, 2000: 173)

Although public health has these powers, they cannot be used in an indiscriminate manner: Governments must demonstrate that the health threat to the community is such that it warrants limiting individual civil liberties (Childress et al., 2002; Dawson & Verweij, 2007; Nuffield Council on Bioethics, 2007; Baylis et al., 2008). In situations where such
infringements are deemed necessary, some public health ethics scholars appeal to the principle of least restrictive or least coercive alternatives for guidance (Upshur, 2002; Childress et al., 2002; Thompson et al., 2006; Baylis et al., 2008). Specifically, if a public health initiative is deemed necessary for the greater good or health of a society yet infringes on the rights and freedoms of individuals, efforts should be made to minimize, to the extent possible, such intrusions before imposing mandatory legislation or other coercive measures (Childress et al., 2002; Upshur, 2002; Nuffield Council on Bioethics, 2007). For example, least restrictive or least coercive alternatives can include “[e]ducation, facilitation, and discussion” (Upshur, 2002: 102). Other examples of least restrictive or coercive alternatives include monitoring, informing and educating, and guiding choices through policy change, incentives, or disincentives (Nuffield Council on Bioethics, 2007: 42). In fact, The Nuffield Council on Bioethics (2007) developed what they call “The Intervention Ladder” to help guide such decision-making for non-clinical public health interventions (see Appendix D for a copy of this ladder).13 Some scholars argue that the more coercive approaches to public health initiatives should be reserved for those instances where “less restrictive measures have failed to achieve appropriate public health ends” (Thompson et al., 2006: 6). In short, a key question in public health ethics analyses, therefore, is what is the justifiable threshold for “the state to introduce programmes that interfere to different degrees in the lives of its population, in order to reduce the risks to the health of all or some of them” (Nuffield Council on Bioethics, 2007: p.15; see also Dawson & Verweij, 2007). Scholars engaged in public health

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13 As a result of my inquiry I suggest in the Future Recommendation section in Chapter 9 that consideration be given to the development of a similar ladder for the clinical public health interventions. I offer a preliminary sketch of what might be included in such a framework in Appendix E.
ethics research offer approaches to achieve this balance through the contribution of new philosophical and analytic frameworks.

**Different Approaches to Public Health Ethics Scholarship**

Scholars have created, advanced, and engaged with public health ethics theory in different ways. Some academics have presented comprehensive philosophical or theoretical conceptualizations of public health ethics (Callahan & Jennings, 2002; Childress *et al.*, 2002; Roberts & Reich, 2002; Baylis *et al.*, 2008). Others have contributed to the development of public health ethics theory through collective engagement with stakeholders involved in public health initiatives (Thompson *et al.*, 2006). Independent researchers, advisory bodies, and public health organizations have created analytic frameworks designed to help public health professionals and other individuals engaged in public health initiatives negotiate and mitigate the associated ethical dimensions and moral tensions of particular public health initiatives (Kass, 2001; European Public Health Ethics Network, 2006; Nuffield Council on Bioethics, 2007; Baum *et al.*, 2007; Tannahill, 2008; Public Health Ontario, 2012; ten Have *et al.*, 2013). Finally, scholars have applied public health ethics lenses to their analyses of specific public health issues such as communicable diseases and environmental health (Upshur, 2002), HIV/AIDS (Nixon, 2006b; Jaffe & Hope, 2010) and global responsibility (Nixon, 2006a; Nixon & Forman, 2008), pandemic planning (Thompson *et al.*, 2006; Baum *et al.*, 2007; Kenny *et al.*, 2010) and general preparedness planning (Swain *et al.*, 2008), obesity (ten Have *et al.*, 2013), and the disclosure of carrier status through NBS (Miller *et al.*, 2009a).
Collectively, existing public health ethics scholarship has generated a host of principles or values deemed integral to the moral framing of public health initiatives. Such key values include the harm principle, least restrictive or coercive alternatives, reciprocity, transparency, accountability, trust, (relational) solidarity, (relational) autonomy, effectiveness, proportionality, and social justice (Upshur, 2002; Childress et al., 2002; Thompson et al., 2003; Benatar, 2003; Krantz et al., 2004; Thompson et al., 2006; Powers & Faden, 2006; Gostin & Powers, 2006; Nuffield Council on Bioethics, 2007; Baylis et al., 2008; Nixon & Forman, 2008; Mascalzoni et al., 2008). These principles and values are then often applied analytically to examine the normative dimensions of the given public health initiative to establish a balance between the rights of the individual and the broader community health goals. Analytic frameworks seem to be particularly useful in this endeavour.

The Use of Frameworks in Public Health Ethics

For more than a decade, scholars engaged in public health practice, policy, and research have focused increasingly on the ethical dimensions of public health initiatives. Scholars internationally have developed frameworks intended to help other academics, health care providers, public health practitioners, and other professionals engaged in public health-related work think through the ethical issues inherent in public health programs, policies, and interventions (Kass, 2001; Baum et al., 2007; Tannahill, 2008; Public Health Ontario, 2012; ten Have et al., 2013). Public health ethics frameworks are analytic tools designed to aid in decision-making by helping professionals engaged in public health related initiatives think through the ethical issues of a given initiative in a systematic and comprehensive manner (Baum et al., 2007; Dawson, 2010; Public Health Ontario, 2012; ten Have et al., 2013).
More specifically, public health ethics frameworks encourage the systematic examination and/or assessment of ethical issues and tradeoffs embedded in existing or future public health programs, policies, or interventions (Baum et al., 2007; Dawson, 2010; Public Health Ontario, 2012; ten Have et al., 2013); facilitate discussion and debate around the perceived goals and objectives, strengths and weakness, benefits and burdens, and ethical challenges of a given public health initiative (Baum et al., 2007; Dawson, 2010; ten Have et al., 2013); enable decision-making consistent with community values (Thompson et al., 2006; Baum et al., 2007; ten Have et al., 2013); and ultimately contribute to the implementation or amendment of public health initiatives that have attempted to minimize or eliminate the ethical tensions (Kass, 2001; Baum et al., 2007; Public Health Ontario, 2012; ten Have et al., 2013).

For frameworks to achieve these goals and objectives, public health ethics scholars have argued for pragmatic frameworks that are general, accessible, acceptable, and flexible (Baum et al., 2007; Dawson, 2010). Public health ethics frameworks can also provide a point of departure for discussion, debate, and deliberation among professionals with multidisciplinary backgrounds and diverse worldviews provided the framework does not emphasize too heavily any one philosophical, theoretical, or conceptual orientation: “If a framework focuses narrowly on one particular perspective, other types of practitioners may not find it useful” (Baum et al., 2007: 661).

Although public health ethics frameworks have many functions and abilities, frameworks do not eliminate disagreements, provide formulaic solutions, or eliminate the need for judgment (Baum et al., 2007; Dawson, 2010; Public Health Ontario, 2012; ten Have et al., 2013).
et al., 2013). Frameworks are not meant to generate “the last word on any topic” (Dawson, 2010: 200). When considering the use of a public health ethics framework for an ethics analysis of a public health initiative, there is no one, single, “right” framework to use (Dawson, 2010). Rather, “[t]he problem to be addressed ought to drive the framework, rather than the other way around” (Dawson, 2010: 200).

In considering the policy and practice of consent for NBS in Ontario I used both a public health ethics lens for my study design, analysis, and interpretation and then conducted an applied public health ethics analysis using a public health ethics framework. In Part II of this chapter I articulate my reasoning behind the use of a public health ethics lens for this inquiry followed by my rationale for selecting Public Health Ontario’s (2012) framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, for the applied component of this inquiry.

**Part II: My Rationale For and Use of a Public Health Ethics Lens for this Inquiry**

*Why I Used a Public Health Ethics Lens for this Inquiry*

Screening and testing programs for an array of health conditions are among the cornerstones of public health (Callahan & Jennings, 2002; Jennings, 2003; Holland, 2007). NBS for PKU and CH is lauded for being among the most significant public health achievements considered invaluable to infant health (American Academy of Pediatrics, 2000; Kwon & Farrell, 2000; Cunningham, 2002). Some scholars insist the quality, efficiency, and comprehensiveness of NBS is facilitated by its public health delivery (Holtzman, 2006).

As previously articulated, public health programs, policies, and interventions introduce distinct challenges: challenges dominated by the inherent competing interests at
play between individuals and the communities within which they live (Beauchamp, 1999; Callahan & Jennings, 2002; Benatar, 2003; Mackie & Sim, 2004; Nixon, 2006a; Dawson & Verweij, 2007; Baum et al., 2007; Dawson, 2010). As with many public health initiatives, while the benefits of a given intervention are often borne by both the individual and society, the potential risks or disadvantages are typically endured by the individual and his or her family alone:

Very often, ethical concerns about public health arise because initiatives and policies are proposed and implemented that can be expected to maintain or improve the health of a target population, but at the expense of some of its individual members. (Holland, 2007: 23)

In considering these tradeoffs within the context of NBS, universal NBS targets the entire infant population to treat a subset of those individuals ultimately affected with one of the disorders on the screening panel. Affected infants are meant to benefit directly by early diagnosis of and treatment for their condition, and society is considered to benefit as well by having healthy, functioning members of society (Faden et al., 1982; Shickle & Chadwick, 1994; Horner, 2000; Schoen et al., 2002).

Most infants and their parents benefit from NBS without becoming intimately connected with their regional NBS program, as the vast majority of infants screen negative.14 A subset of infants and their parents, however, bear the brunt of what some might argue constitute the potential negative implications of NBS (such as, “unnecessary anxiety, stigma, false reassurance or side effects from the procedure itself” (Horner, 2000: 49)) for the overall effectiveness of the program and ultimately for the health of the community (Shickle & Chadwick, 1994; Horner, 2000). Examples of parents who might be perceived to bear an

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14 I use the language of benefit, yet recognize the possibility that a negative screen could potentially foster false reassurance, as false negatives are a possibility in any screening program (Horner, 2000).
unwanted burden for the greater good of society include those parents whose infants receive false positive screens (Shickle & Chadwick, 1994; Waisbren et al., 2003; Hewlett & Waisbren, 2006; Wilcken, 2010) and those whose infants screened positive and, subsequent to follow-up testing, receive ambiguous and inconclusive diagnoses, rendering infants “patients-in-waiting” (Timmermans & Buchbinder, 2010). Other examples might include parents who receive information about their infants’ carrier status (information that they otherwise would not have wanted to receive) (Miller et al., 2009a), as well as parents who would not have wanted their infants’ blood spots stored for future uses (Rothwell et al., 2010; Tarini et al., 2010).

Considering NBS in Ontario specifically, NBS had been an explicitly public health service in Ontario for 40 years until the program expanded in 2006. Although the NBS program is no longer affiliated with the Public Health Laboratory, Newborn Screening Ontario has since underscored its commitment to preserving NBS’s public health roots (Honeywell & Cloutier, 2007). Furthermore, scholars engaged in research involving Ontario’s NBS program continue to frame it as a public health program (Bombard et al., 2012; Miller et al., 2010b; Araia et al., 2012). The history of NBS as a public health program; the interconnectedness between the health of individuals and society and the associated tensions that it can bring within the context of NBS; Newborn Screening Ontario’s commitment to preserving the public health dimension of NBS; and the sustained framing of Ontario’s NBS program as a public health program worked collectively to legitimate my decision to design my inquiry into stakeholder perceptions of ethical issues embedded in

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15 It is important to note that in Ontario, parents must expressly consent to receiving carrier status results by discussing first with their health care provider and then contacting Newborn Screening Ontario to receive the results (Newborn Screening Ontario, n.d.j).
NBS in Ontario and their attitudes towards consent for NBS in particular through a public health ethics lens.

In addition, my data solidified the appropriateness of a public health ethics lens for my analysis and interpretation. Given the iterative nature of qualitative inquiry, whereby the theory informs the data and the data informs the theory (Sandelowski, 1993), the evolution of my conceptual framework is necessarily dependent not only on my study design, but also on my approaches to data collection, analysis, and interpretation, all of which will be explained in subsequent chapters of this dissertation. However, since my conceptual framework is dependent, in part, on my findings, I introduce at this juncture my decision to focus my analysis and interpretation on consent; define the four salient public health ethics principles that I identified in my data on consent and explain why they are useful principles to consider in relation to explorations of consent within a public health context; and, finally, to provide the rationale behind my decision to then apply these principles to an analytic public health ethics framework.

My findings revealed a focus on perceptions of and attitudes towards both the current and possible future approach to consent for NBS in the province (implied consent and express consent respectively). The policy and practice of consent for a public health initiative is a topic of significant debate in public health ethics scholarship, as it necessarily invokes discussion about the appropriate emphasis that should be placed on the principle of autonomy within a public health context (Holland, 2007; Dawson, 2010). As explained earlier in this chapter, some public health ethics scholars argue that the act of falling on the side of individual liberty, expressed through consent, is to merely redefine the dilemma rather
than to reach a resolution (Horner, 2000; Holland, 2007), whereas others maintain that in a liberal democracy such as Canada, preserving individual liberty with respect to choices in healthcare is an inviolable civil right and must be considered seriously when addressing the tensions between individual rights and freedoms and population health (Gostin, 2003; Nixon & Forman, 2008). Given the polar public health ethics perspectives on this topic, I wanted to analyze and interpret my data using a public health ethics lens that could provide new insights in the continued effort to find a balance between the two poles on this highly debated issue.

As I explained in Part I of this chapter, there is no one public health “ethic.” Yet, a common theme that runs through public health ethics scholarship is an exploration and application of key principles and values deemed quintessential to public health (Kass, 2001; Childress et al., 2002; Upshur, 2002; Thompson et al., 2006; Miller et al., 2009a; ten Have et al., 2010; Lee, 2012). My public health ethics-informed analysis and interpretation of perceptions of and attitudes towards the policy and practice of consent for NBS in Ontario led me to identify in my data the following four public health ethics principles: least restrictive alternatives, effectiveness, autonomy, and social justice. I explain each of these principles in greater detail below, as these principles also informed the lens through which I reviewed the literature in the chapter that follows (see Chapter 3).
The Four Salient Public Health Ethics Principles Most Prevalent for this Inquiry

1. **The principle of least restrictive alternatives: consent as a least restrictive alternative for clinical public health initiatives**

   The principle of least restrictive or least coercive alternatives underscores the argument that while there may be a number of different ways to achieve public health goals and objectives, the least restrictive alternative should be implemented whenever possible:

   This principle recognizes that a variety of means exist to achieve public health ends, but that the full force of state authority and power should be reserved for exceptional circumstances and that more coercive methods should be employed only when less coercive methods have failed. Education, facilitation, and discussion should precede interdiction, regulation or incarceration. (Upshur, 2002: 102)

   Upshur (2002) underscores “[e]ducation, facilitation, and discussion” (p.102) as among the least restrictive approaches that can be used to achieve public health goals. The Nuffield Council on Bioethics (2007) created “The Intervention Ladder” to capture the gradation of possible public health approaches from least intrusive to most intrusive approaches for non-clinical public health interventions (see Appendix D: The Nuffield Council on Bioethics’ “Intervention Ladder”). Public Health Ontario’s ethics framework, designed initially for public health research initiatives (2012), proposes alternatives to obtaining individual informed consent in those instances where seeking individual informed consent is not “required, feasible, or appropriate” (p.4). Such alternatives include, “broad consent, opt-out, and consultation with a representative sample of the population of interest” (p.4).

   In considering public health’s clinical interventions (e.g., NBS, vaccination), a similar gradation of intervention exists: “express consent” epitomizing the least restrictive or least coercive of the options and “mandatory interventions without opt-out provisions” representing the most restrictive or coercive approaches. I will argue throughout this
dissertation that the practice of informed consent – both implied and express consent – are
two examples of least restrictive alternatives (although implied consent would be considered
more restrictive than express consent) that can be among the approaches implemented for a
given public health program, policy, or intervention. (See Appendix E: Preliminary
framework for thinking through the spectrum of autonomy within the context of medical
public health interventions.)

2. The principle of effectiveness: examined within a context of
least restrictive alternatives

While public health ethics scholars tend to make general appeals to implement least
restrictive or coercive alternatives whenever possible, less attention is paid to the potential
moral concerns, questions of effectiveness, and potential for harm that these least restrictive
options can introduce (Macintyre & Petticrew, 2000; Guttman & Salmon, 2004; Nuffield
Council on Bioethics, 2007; Faden & Shebaya, 2010). Referencing the Nuffield Council’s
“intervention ladder” as an example, Faden and Shebaya (2010) argue,

Continua of this sort also oversimplify the complex impact of interventions on choice and
liberty and on relations between citizens and the state. Incentives are not always less
restrictive of choice than disincentives, and health promotion campaigns, which are
generally ranked at or near the least intrusive end of the continuum, are not always
without significant moral concern. Ad campaigns that are transparently sponsored by
public health agencies to prevent transmission of influenza by promoting personal
infection control practices or reduce obesity by encouraging exercise and healthy eating
do not raise the same moral issues as the embedding of anti-drug or abstinence messages
in the story lines of entertainment television programming by these same authorities
(FCC 2000; Forbes 2000 (Other Internet Resources); Goodman 2006; Krauthammer
2000; Kurtz and Waxman 2000). While the latter poses important questions about respect
for liberty, government over-reaching and democratic legitimacy, the limited
effectiveness of many ad campaigns raises important questions about whether the state is
underserving its public health mission. Moreover, in the case of public health problems
like obesity, a reliance on health promotion campaigns and other strategies focused on
influencing the behavior of individuals may fail to place appropriate burden on the
corporate interests and structural social inequalities that arguably account for much of the problem. Thus, depending on the circumstances, health promotion campaigns may be unjust as well as ineffective (Buchanan 2008; Crawford 1998; Faden 1987; McLeroy, Bibeau, Steckler, & Glanz 1988). (p.17-18)

Similarly, other less restrictive alternatives can be equally unjust and ineffective, depending on the circumstances, and can potentially cause harm:

The implications of this observation are that well-intentioned and plausible interventions, even of a non-invasive kind involving only education, can do unanticipated harm. This suggests that there is a duty on those introducing such measures to monitor their actual impact over appropriate timeframes, rather than simply assuming they are beneficial. (Nuffield Council on Bioethics, 2007: 32)

Tannahill (2008) argues a similar point in his appeal to the role of theory in helping anticipate “potentially hidden harm[s]” (p.388):

[T]heory has a part to play in identifying possible harms and actions to mitigate them, and all the more so given that a further challenge for evaluation is to pay more attention to detecting potentially hidden harm (for example to wellbeing, or confined to certain population groups). (Tannahill, 2008: 388)

In the chapters that follow, I will explore the challenge of moral justification and effectiveness within the context of participant perceptions of and attitudes towards implied and express consent for NBS in Ontario.

To explore this principle within the context of my data on consent, I have drawn on Childress et al.’s (2002) definition of effectiveness to mean the extent to which the policy implemented achieves its desired goals:

It is essential to show that infringing one or more general moral considerations will probably protect public health. For instance, a policy that infringes one of more general moral considerations in the name of public health but has little chance of realizing its goal is ethically unjustified. (Childress et al., 2002: 173)

The current definition of effectiveness is framed in a way to underscore that the “burden of moral proof” (Childress et al., 2002: 173) lies with public health practitioners and policy
makers to ensure the ethical justification of the program, policy, or intervention in question by demonstrating its ability to deliver on its population health promises (Kass, 2001; Childress et al., 2002; Baylis et al., 2008; Public Health Ontario, 2012). Within my study, I have similarly applied the public health principle of effectiveness to the policy and practice of consent. Specifically, this approach examines perceptions of whether a consent policy designed and implemented within the context of NBS to protect against infringements on individual liberty, yet potentially at the “expense” of population health, achieves (in the case of implied consent) or could achieve (in the case of express consent) its objectives in facilitating informed, autonomous decision-making. A least restrictive alternative within a public health context (as examined through a public health ethics lens that strives to achieve a balance between individual and community tensions) must arguably succeed both in terms of its promise to maximize the potential for self-governance while also achieving its broader population health goals. In my public health ethics analysis of consent, both the principle of effectiveness and the principle of autonomy work together to explore perceptions around the extent to which consent in practice meets the ethical concept’s ideals and objectives.

3. The principle of autonomy: a debated principle in public health ethics

In contemporary liberal democracies such as Canada, individual liberty – the foundation upon which the principle of autonomy and self-determination is based – is extremely important in law, public deliberation, and policy-making (Wildeman & Downie, 2001; Thompson et al., 2006; Nuffield Council on Bioethics, 2007; Public Health Ontario, 2012). The ethical concept of consent is “grounded in the principle of respect for autonomy” (Holland, 2007: ix) and implemented in health care contexts specifically to
protect against what the Nuffield Council on Bioethics (2007) has labelled “the erosion of individual freedom” (p.42). Although the principle of autonomy is paramount in clinical practice and medical research contexts, some public health ethics scholars argue that autonomy is an inappropriate emphasis for public health (O’Neill, 2002; Dawson & Verweij, 2007; Holland, 2007; Baum et al., 2007; Baylis et al., 2008; Dawson, 2010). Horner (2000) insists that privileging autonomy within public health contexts merely redefines the dilemma between the rights and freedoms of the individual versus broader community goods rather than offering a solution: “This relentless drive towards autonomy takes us to the other end of our dilemma, from that of utilitarianism and, as such, does not resolve it” (p. 50). Holland (2007) supports Horner’s (2000) argument, urging against the tendency to privilege one principle over another:

[T]he central ethical dilemma in public health arises precisely when we feel compelled to restrict individuals’ autonomy in the interests of the community. So, invoking the principle of autonomy amounts to simply opting for the individual against the community, which is clearly no way to resolve the dilemma. (Holland, 2007: 27)

For these reasons Holland (2007) maintains that “the principle of autonomy is not going to be much use in public health ethics” (p.27).

I would argue, however, that considering the principle of autonomy within the broader public health context is critical when examining least restrictive alternatives for their ability to achieve the sought-after equilibrium between liberty and self-governance on the one hand and state intervention and infringement on the other. For the extent to which an approach can be deemed least restrictive depends largely on its ability to protect individual rights and freedoms without sacrificing the larger public health goals. Three additional reasons in support of considering the principle of autonomy within the context of public
health and public health ethics analyses include the fact that individual liberty is constitutionally protected within the context of healthcare, that the principle of autonomy is built into public health ethics frameworks, and that scholars debate whether parents should consent to such clinical public health interventions as NBS.

First, as mentioned earlier in this chapter, countries in the Western world are liberal democracies (Nuffield Council on Bioethics, 2007; Holland, 2007). Consequently, individual liberty with respect to health care decision-making is constitutionally protected, albeit with limitations (Wildeman & Downie, 2001; Nuffield Council on Bioethics, 2007). Second, given that individual liberty is constitutionally protected in many jurisdictions, including Canada, scholars have begun to integrate the principle of autonomy and the concept of consent into the very infrastructure of their public health ethics frameworks. Kass’ (2001) seminal public health ethics framework encourages users to consider “risks to liberty and self-determination” while reflecting among “the known or potential burdens of the program” (p.1779). A host of other public health ethics frameworks have similarly incorporated informed consent, informed choice, liberty, autonomy, and/or self-determination into their analytic frameworks as well (see Baum et al., 2007; Swain et al., 2008; Petrini & Gainotti, 2008; Public Health Ontario, 2012; ten Have et al., 2013). Public Health Ontario (2012), for example, adopted a question-based approach for its framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, and devoted an entire question to consent: “Is individual informed consent warranted? Is it feasible? Is it appropriate? Is it sufficient?” (p.4); thus raising the question of when consent is and is not warranted in a public health context. The presence of this ethics principle in public health ethics
frameworks arguably strengthens the argument that while issues of autonomy as expressed through informed consent may not dominate as they do in medical and research ethics, they are nonetheless important considerations when examining and assessing public health programs, policies, and interventions. As Baylis et al. (2008) argue, while autonomy “looms especially large in clinical and research ethics where the primary target of intervention is a specific—and vulnerable—patient and/or research participant, it is also a value that must be present in public health initiatives” (p.201). Finally, the third reason in support of the importance of considering autonomy in public health ethics is that public health programs such as NBS have found themselves engaged in debates as to whether express consent for NBS – and expanded NBS in particular – should be implemented (Wildeman & Downie, 2001; Pollitt, 2004; Newson, 2006; Nijsingh, 2007; Tarini et al., 2008; Harrell, 2009; Ross, 2010). For these reasons the principle of autonomy, within the clinical public healthcare context, cannot be eliminated from normative public health ethics analyses. While scholars who insist on privileging autonomy and self-determination over other public health ethics principles arguably do little to achieve a balance between individual liberties and the welfare of the community, to dismiss the principle altogether would also be misguided.

4. **The principle of social justice: considering social justice within the context of consent**

An individual’s ability to make true, autonomous decisions can, however, depend – at least in part – on their social situation. In particular, the extent to which one is negatively impacted by the social determinants of health can work to undermine one’s autonomy and ability to self-govern:
[A] public health lens brings into focus forms of disadvantage such as the impact of social determinants that may affect autonomy. Poverty, for example, may reduce choice or the opportunity to express a preference. It may even affect the perception that an individual has a choice. Consequently, persons with limited means may, for example, find it difficult to participate in community forums. (Public Health Ontario, 2012: 8-9)

A social determinant of health (e.g., social class, occupation, income, and education) is a “socially controllable factor outside the traditional health care system that is an independent partial cause of an individual’s health status” (Sreenivasan, 2008: 2). While such factors as socioeconomic status, education, and employment can have an impact on one’s health, some scholars argue that health care itself (both “personal medical care and public health”) is also a crucial social determinant of health (Sreenivasan, 2008: 2). Given the impact of social determinants of health on individual and group health, many scholars consider the principle of social justice as the bedrock of ethically sound public health programs, policies, and interventions (Beauchamp, 1999; Gostin & Powers, 2006; Powers & Faden, 2006; Baylis et al., 2008; Faden & Shebaya, 2010):

[Public health based on social justice gives rise to important policy imperatives such as improving the public health system, reducing socioeconomic disparities, addressing health determinants, and planning for health emergencies with an eye for the most vulnerable. (Gostin & Powers, 2006: 1054)

Scholars committed to incorporating a social justice lens in public health and health policy have consistently embedded the principle of social justice in public health ethics theory and frameworks (Kass, 2001; Gostin & Powers, 2006; Powers & Faden, 2006; Baum et al., 2007; Baylis et al., 2008; Public Health Ontario, 2012). Specifically, appeals to social justice in this body of literature tend to focus on “the twin moral impulses that animate public health: to advance human well-being by improving health and to do so by focusing on the needs of the most disadvantaged” (Gostin & Powers, 2006: 1054). Some public health ethics
scholars call for the implementation of public health initiatives that are sensitive to the social determinants of health and, consequently, attempt to ensure a fair distribution of associated benefits and burdens in an effort to achieve health equality and equity (Kass, 2001; Gostin & Powers, 2006; Baum et al., 2007; ten Have et al., 2013). Being attentive to issues of social justice within the context of public health programs, policies, and interventions means, at least in part, an effort to rectify “patterns of systematic disadvantage that undermine the well-being of people whose prospects for good health are so limited that their life choices are not even remotely like those of others” (Gostin & Powers, 2006: 1054).

Powers and Faden (2006) contribute to this scholarship on social justice and public health by proposing a theory of social justice rooted in the concept of well-being. Well-being is a critical dimension of the human experience which Powers and Faden (2006) argue is not captured under purely distributive understandings of justice:

Having the respect of others as social equals, having personal attachments, and having the capacity for leading self-determining lives are essential elements of human well-being but they are not securable simply by ensuring a just distribution of resources such as wealth and income. Rather, they require attention to the structure of a wide range of social institutions and practices that govern far more than how desirable goods such as income or wealth are distributed. (p.192)

These scholars argue for an approach to social justice that considers

the morally salient human ends that each of us want regardless of whatever else we might want. For us, these ends include the respect of others, personal security, health, the development of reasoning capacities and of capacities for attachment to others, and to determine for oneself some important aspects of one’s own destiny. (Powers & Faden, 2006: 191)

To this end, Powers and Faden (2006) articulate a comprehensive interpretation of well-being that encompasses these six facets of well-being: health, personal security, reasoning, respect, attachment, and self-determination. For Powers and Faden (2006), these six facets of well-
being are “each distinctive human ends, all of which matter to justice generally as well as to justice in the specific contexts of public health and health policy” (p.191).

Although each dimension of well-being is distinctive, they are interconnected with each other and with the social determinants of health more broadly: inequalities in one area of life ultimately have implications for other areas and, as a result, can contribute to the perpetuation of systematic disadvantage (Gostin & Powers, 2006; Powers & Faden, 2006; Baylis et al., 2008). Respect, for example, is a critical component of well-being and particularly salient when considering health disparities: “...the members of the disadvantaged group are accorded less respect, which frequently translates into reduced self-respect, reduced expectations, and reduced capacity for self-determination” (Powers & Faden, 2006: 88). Powers and Faden (2006) argue that health policy makers have an obligation to help cultivate each of the six dimensions of well-being to ensure, to the extent possible, that individuals achieve “a sufficient amount of each of the essential dimensions of well-being” in an effort to redress social injustices:

A commitment to social justice, as we explicate it, attaches a special moral urgency to remediating the conditions of those whose prospects are poor across multiple dimensions of well-being. Placing a priority on those so situated is a hallmark of public health. (p. 82)

I included an exploration of the principle of social justice in my public health ethics analysis of consent for NBS because social justice is a critical principle in many public health ethics analyses, and because a number of my participants introduced social determinants of health when discussing their perceptions of and attitudes towards consent for NBS. I decided to draw on Powers and Faden’s (2006) theory of social justice with their emphasis on well-being not only because of the intersection between social determinants of health and well-
being, but also because social goods such as respect, self-determination, and reasoning, for example, are firmly embedded in the theory and practice of consent. In the chapters that follow, I suggest that a public health ethics analysis of consent offers an opportunity to examine the possible ways in which consent policy and practice can either ameliorate or perpetuate health injustices for individuals and groups. Public health has an obligation to “evaluate the impact of its policies and practices, not only on health, but on all the dimensions of well-being” as each dimension “identifies a separate kind of injustice” (Powers & Faden, 2006: 83). By interpreting well-being through a multi-faceted lens, when considering the social justice components of a given public health policy or practice like consent, the extent to which the public health initiative compromises or strengthens each dimension of well-being can be used as moral arguments either for or against the policy or practice in question (Powers & Faden, 2006). A public health ethics analysis of consent offers an opportunity to identify the moral strengths and potential moral weaknesses of consent in practice. In addition, a public health ethics lens encourages professionals engaged in public health initiatives to consider the ways in which consent can be implemented to engage parents with varying capacities in an effort to elevate, rather than diminish, well-being and in so doing contribute to the reduction of health inequities.

An Applied Analysis of Consent Using a Public Health Ethics Framework

The decision to apply my findings to a public health ethics framework stemmed from the analysis and interpretation of my data. Specifically, many of my participants perceived informed consent for NBS – both implied and express consent – through a critical lens identifying perceived benefits and burdens of existing (implied consent) and hypothetical
(express consent) consent policy for NBS in Ontario (see Chapters 6 & 7). Examining these perceived benefits and burdens through a public health ethics lens led me to frame them within the context of the broader public health ethics principles of least restrictive alternatives, effectiveness, autonomy, and social justice. This framing of consent illuminated many participants’ uncertainty as to whether consent could successfully protect individual rights and freedoms without compromising the broader public health goals of the NBS program. Given the highlighted tensions associated with consent for NBS, I decided to apply the policy and practice of consent for NBS to an analytic public health ethics framework crafted and honed specifically to facilitate a comprehensive and systematic exploration of the unique ethical challenges introduced through public health programs, policies, and interventions. Applying the policy and practice of consent for a public health program such as NBS to a public health ethics framework allows for a systematic assessment of the policy and practice; provides a comprehensive structure to identify potential benefits and burdens; and creates an opportunity either to improve an existing policy and practice or to mitigate potential harms as a preemptive strategy in advance of implementation (Kass, 2001; Baum et al., 2007; Tannahill, 2008; Public Health Ontario, 2012; ten Have et al., 2013).

In considering the possible public health ethics frameworks for this applied public health ethics analysis, I focused specifically on general analytic frameworks that could aid current and future NBS advisory committees in Ontario think through the issue of consent in a comprehensive way. Applying the policy and practice of consent to a public health ethics framework and challenging some of the taken-for-granted assumptions embedded in this practice will hopefully help guide future discussion and debate regarding consent for NBS in
Ontario in a way that achieves the public health ethics balance of protecting both individual rights and population health.

Why I Chose to Use Public Health Ontario’s Public Health Ethics Framework

In selecting a framework for this applied ethics analysis of my examination of stakeholder perceptions of and attitudes towards consent for NBS I focused on those public health ethics frameworks that provided general analytic tools in the form of a question-answer format designed specifically to facilitate a guided discussion and to aid in decision-making. As explained in Part I of this chapter, there is no “one” public health ethic or one “right” public health ethics framework applicable to all public health programs, policies, or interventions (Dawson, 2010). When considering the use of an analytic framework Dawson (2010) asserts that “[t]he problem to be addressed ought to drive the framework, rather than the other way around” (p.200). Public health ethics frameworks are meant to serve as guides, rather than producing “the last word on any topic” (Dawson, 2010: 200). Among the strengths of these deliberative aids is that they are iterative and “provisional and heuristic” in nature (Dawson, 2010: 200). Other characteristics that constitute a strong analytic public health ethics framework include the extent to which the framework is pragmatic, accessible, general, acceptable, and flexible (Baum et al., 2007; Dawson, 2010). The range of public health ethics values reflected in the framework is also an important criterion to consider when selecting a framework for a public health ethics analysis (Dawson, 2010).

With these criteria in mind, I narrowed the selection of frameworks first according to their practical applicability, accessibility, and range of public health ethics values. Since my analysis and interpretation of participant perceptions towards consent for NBS introduced
such public health ethics principles as least restrictive alternatives, effectiveness, autonomy, and social justice, I wanted to use a public health ethics framework that encompassed a number of public health ethics principles rather than a framework that focused on one organizing principle such as social justice, for example. In addition, I wanted to select a framework that would be both practical and accessible. I interpreted pragmatic and accessible frameworks as those designed in a user-friendly manner with accompanied instruction on how to help individuals engaged in public health related work think through public health ethics principles and issues raised by a given public health program, policy, or intervention (Baum et al., 2007; ten Have et al., 2013). I identified four pragmatic frameworks that included numerous public health ethics values and presented them for consideration and deliberation in a clear, constructive, accessible way using a guided question format: Kass (2001), Baum et al. (2007), Public Health Ontario (2012), ten Have et al. (2013) (see Table 1).16

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16 One other framework by Swain et al. (2008) offered a guided eight-question approach, but the first six questions were from Kass’ (2001) framework and there was no guidance in terms of how professionals should use the framework. Also, the two additional questions incorporated into Swain et al.’s (2008) framework were captured in the other frameworks and, therefore, I did not pursue Swain et al.’s (2008) framework for my analysis.
**Table 1: Public Health Ethics Frameworks Comparison Chart**

This colour coded public health ethics framework comparison chart highlights the large degree of overlap in terms of the public health values identified as important for consideration.

<table>
<thead>
<tr>
<th>KEY: Analytic Framework Themes</th>
<th>Goals and objectives of public health program</th>
<th>Effectiveness</th>
<th>Benefits</th>
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<tr>
<td></td>
<td>Goals and objectives of public health program</td>
<td>Burdens</td>
<td>Community engagement</td>
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<td></td>
<td>Effectiveness</td>
<td>Balance of benefits and burdens</td>
<td>Autonomy</td>
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<td>Benefits</td>
<td>Social justice</td>
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<tr>
<th>Kass, 2001</th>
<th>Baum et al., 2007</th>
<th>ten Have et al., 2013</th>
<th>Public Health Ontario, 2012</th>
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<tr>
<td>What are the public health goals of the proposed program?</td>
<td>Determine population-level utility of the proposed action</td>
<td>How does the programme affect physical health?</td>
<td>What are the objectives of the initiative? How are they linked to potential improvements in public health?</td>
</tr>
<tr>
<td>How effective is the program in achieving its stated goals?</td>
<td>Demonstrate evidence of need and effectiveness of actions</td>
<td>How does the programme affect psychosocial well-being?</td>
<td>Can the objectives be achieved using the proposed methods?</td>
</tr>
<tr>
<td>What are the known or potential burdens of the program?</td>
<td>Establish fairness of goals and proposed implementation strategies</td>
<td>How does the programme affect equality?</td>
<td>Who are the expected beneficiaries of the knowledge gained or other benefits?</td>
</tr>
<tr>
<td>Can burdens be minimized? Are there alternative approaches?</td>
<td>Demonstrate accountability</td>
<td>How does the programme affect informed choice?</td>
<td>What are the burdens and potential harms associated with the proposed initiative? Who bears them?</td>
</tr>
<tr>
<td>Is the program implemented fairly?</td>
<td>Assess expected efficiencies and costs associated with the proposed action</td>
<td>How does the programme affect social and cultural values?</td>
<td>Are burdens and potential harms justified in light of the potential benefits to participants and/or to society?</td>
</tr>
<tr>
<td>How can the benefits and burdens of a program be fairly balanced?</td>
<td>Consider political feasibility and community acceptance</td>
<td>How does the programme affect privacy?</td>
<td>Is selection of participants fair and appropriate?</td>
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<td>“Finally, several other principles that have historically been central in the development of the field of bioethics – autonomy, non-maleficence, and beneficence – have a role in public health, but may be relevant to a lesser degree than they have been in bioethics.” (p. 663)</td>
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<td>Is individual informed consent warranted? Is it feasible? Is it appropriate? Is it sufficient?</td>
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<td>Is community engagement warranted? Is it feasible? What level of engagement is appropriate?</td>
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<td>What are the social justice implications of this initiative?</td>
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<td>What are the potential longer-term consequences?</td>
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I also wanted to use a framework that was general in scope; one that would be applicable to a host of public health initiatives on a diverse range of topics—and for my particular issue at hand, a framework that would facilitate an examination of the policy and practice of consent for NBS. Of the four frameworks presented in the framework comparison table above, ten Have et al.’s (2013) framework was purposefully developed within the context of obesity and overweight literature and geared towards professionals engaged in public health initiatives pertaining to obesity and overweight work. Given the specific focus of this framework I did not pursue ten Have et al.’s (2013) framework for my applied analysis of consent. Public Health Ontario’s (2012) framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, was designed initially to help scholars, researchers, and professionals engaged in public health research or evidence-generating initiatives think through the ethical issues using a public health ethics lens rooted in the tenets of public health. Given the research focus, this framework takes as its point of departure the Canadian *Tri-Council Policy Statement 2, Ethical Conduct for Research Involving Humans* with a specific public health ethics focus on its three core principles: “respect for persons, concern for welfare, and justice” (Public Health Ontario, 2012: 2). These principles were “examined with an emphasis on the inter-relatedness of the welfare of individuals and communities and a positive obligation to promote equity and reciprocity” (Public Health Ontario, 2012: 2). This normative framing is not, I would argue, unique to public health research initiatives, but rather encapsulates the goals and objectives of public health more generally. Moreover, while the framework questions were designed specifically for evidence-generating initiatives, the principles upon which they were based
are rooted in broad conceptualizations of the public health ethics principles of autonomy, social justice, and the interconnectedness of individuals and the community particularly as it pertains to health. In addition, the accompanying instructional on how to apply the framework is general in nature. Therefore, I maintain this framework has a generality and flexibility in its applicability that extends beyond research generating public health initiatives. Consequently, I considered this framework further in relation to the other criteria necessary when selecting a public health ethics framework.

The acceptability of a given framework is another characteristic to consider when contemplating the selection of a framework (Baum et al., 2007; Dawson, 2010). This consideration ultimately led me to select Public Health Ontario’s (2012) framework for my analysis. Baum et al. (2007) underscore the importance of implementing a framework that can appeal to professionals with multidisciplinary backgrounds and diverse worldviews. In addition, scholars have found that the cultural and political backdrop of a given society can influence how ethical tensions in public health programs, policies, and interventions are negotiated, particularly with respect to achieving a balance between protecting the rights and freedoms of individuals and protecting the health of society (European Public Health Ethics Network, 2006; Tannahill, 2008; ten have et al., 2010). A final contributing factor to the acceptability of a framework is determined by professionals engaged in public health work who apply the framework to determine its usefulness and to identify any obstacles to implementation (Baum et al., 2007; ten Have et al., 2013). In examining the applicability of the frameworks, three reasons solidified my decision to use Public Health Ontario’s analytic framework for my analysis.
First, the working group that developed this public health ethics framework, comprised of professionals with expertise in public health ethics, come from different philosophical and theoretical perspectives, arguing from shared understandings rather than a single moral theory. That they all endorse the normative principles put forth in this framework suggests it is a model for public health ethics analysis that will appeal to a broad range of professionals engaged in public health endeavours.

Second, this public health ethics framework evolved through what the working group members have labelled a consultation process (Public Health Ontario, 2012). Earlier iterations of this framework were distributed to public health practitioners and ethicists for feedback. A full draft of this framework was circulated in June 2011 “to public health units across Ontario, independent academics working in the area of public health and ethics, and individuals at the Canadian Institutes of Health Research, the Public Health Agency of Canada, the Panel on Research Ethics, the Registered Nurses Association of Ontario, and the U.S. Centres [sic] for Disease Control” for comment (Public Health Ontario, 2012: 1). Comprehensive feedback from these individuals and institutions was incorporated into the 2012 version of the framework that I have used for my analysis. That practitioners, scholars, and other professional groups and institutions provided feedback and that the framework was adjusted to reflect that feedback speaks to the increased likelihood of acceptability of this framework in Ontario specifically and North America more broadly.

Finally, given that culture and politics were found to contribute to how competing tensions between individuals and communities are addressed (European Public Health Ethics
Network, 2006), using a framework developed in the province where it will be applied may have advantages in terms of the values and principles privileged.

The final characteristic of a strong analytic public health ethics framework is flexibility. Public Health Ontario’s (2012) framework explicitly acknowledges the flexibility of frameworks and the recognition that changes to the framework will be necessary as the framework is used in the assessment of ethical issues embedded in a range of public health programs, policies, and interventions:

As with all frameworks, the guiding questions are not algorithms to provide the “right” answer. They are meant to help with the systematic examination of the issues, including appropriate consideration of the interests of all stakeholders. In addition, as the questions are applied to more and more cases, we anticipate refinements to the questions and interpretive text. We encourage any users of this framework to share with us any refinements they introduce, in the spirit of continual improvement of this document. (Public Health Ontario, 2012: 2)

In short, Public Health Ontario’s (2012) framework embodies all of the characteristics of a strong analytic framework and, therefore, renders it a solid and sensible point of departure for an exploration of perceptions of consent policy and practice for NBS in Ontario.

**Modifications to the Framework**

The explicit flexibility of Public Health Ontario’s (2012) framework coupled with Dawson’s (2010) philosophy that the topic of inquiry should guide the framework rather than forcing the framework onto the issue supports the slight modifications I made to the framework to aid in my public health ethics analysis of the policy and practice of consent for NBS. The original purpose of this framework was to help public health professionals engaged in research generating initiatives think through public health ethics issues connected
to their work. In order to extend the framework’s applicability to a wider array of public health undertakings and policies rather than circumscribing its use to public health research initiatives, I modified a few of the questions to render them applicable to public health programs, interventions, policies, and practices (see Table 2).

Specifically, I made slight changes to four of the ten questions. The first change I made was to question number two. The original question from Public Health Ontario’s framework asked, “Can the objectives be achieved using the proposed methods?” Given the research specific target of this framework, “methods” refer explicitly to study design and methodological rigour (Public Health Ontario, 2012: 3). I modified this question and changed “method” to “approach” to reflect the perceived need of my participants to find new ways of obtaining informed consent.

I also made a change to question three. The original question 3 in this framework reads, “Who are the expected beneficiaries of the knowledge gained or other benefits?” (Public Health Ontario, 2012: 13). I modified the question to be more suitable for public health ethics analyses of a wider array of public health undertakings, replacing “knowledge gained or other benefits” with “program, intervention, policy, or practice”: “Who are the expected beneficiaries of the program, intervention, policy, or practice?”

I also modified question five. The original question read, “Are burdens and potential harms justified in light of the potential benefits to participants and/or society?” (Public Health Ontario, 2012: 14). I changed “participants” to “individuals” to accommodate the use of this framework for the exploration of other public health initiatives such as programs,

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17 For a list of the original questions see Table 1 Column 4.
interventions, policies and practices. I made a similar change in question six to reflect the focus on individuals generally rather than a research focus on participants. All of the other questions remained the same.

Table 2. A Modified* Framework for the Ethical Conduct of Public Health Initiatives

<table>
<thead>
<tr>
<th>Question #</th>
<th>Questions</th>
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<tbody>
<tr>
<td>1</td>
<td>What are the objectives of the initiative? How are they linked to potential improvements in public health?</td>
</tr>
<tr>
<td>2</td>
<td><strong>Can the objectives be achieved using the proposed approach?</strong></td>
</tr>
<tr>
<td>3</td>
<td>Who are the expected beneficiaries of the program, intervention, policy, or practice?</td>
</tr>
<tr>
<td>4</td>
<td>What are the burdens and potential harms associated with the proposed initiative? Who bears them?</td>
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<tr>
<td>5</td>
<td><strong>Are burdens and potential harms justified in light of the potential benefits to individuals and/or to society?</strong></td>
</tr>
<tr>
<td>6</td>
<td><strong>Is the selection of individuals fair and appropriate?</strong></td>
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<tr>
<td>7</td>
<td>Is individual informed consent warranted? Is it feasible? Is it appropriate? Is it sufficient?</td>
</tr>
<tr>
<td>8</td>
<td>Is community engagement warranted? Is it feasible? What level of engagement is appropriate?</td>
</tr>
<tr>
<td>9</td>
<td>What are the social justice implications of this initiative?</td>
</tr>
<tr>
<td>10</td>
<td>What are the potential longer-term consequences?</td>
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</table>

*Questions in bold font reflect my modifications. See Table 1 column 4 on p.46 for the original framework questions (Public Health Ontario, 2012).

I have used this modified public health ethics framework with the purpose of asking new questions of the policy and practice of informed consent within the context of NBS in Ontario to aid in ongoing advisory committee deliberations on this issue. Challenges to express consent are not new and not unique to NBS. However, by framing consent policy as a practice that can be evaluated and then applying Public Health Ontario’s public health ethics framework to my findings, allows for a thorough examination of the perceived benefits and risks of the policy and practice of express consent within a public health context. In addition, such an applied analysis challenges the presumed good often attributed to such purported liberty-preserving mechanisms as express consent.
**Chapter Conclusion**

This two-pronged approach to my public health ethics inquiry – the identification of relevant public health ethics principles followed by the use of a specific analytic tool designed for public health initiatives – applied to perceptions of the policy and practice of consent for NBS in Ontario enables an opportunity to think critically about tensions between individual liberties and social welfare in public health. The public health ethics principles outlined in this chapter recur throughout the chapters that follow, including the literature review (see next chapter, Chapter 3). While the vast majority of scholarship on NBS and consent that I examine in my literature review was not conducted through a public health ethics lens specifically, many of the findings reported arguably resonate with the public health ethics principles I have identified (e.g., least restrictive alternatives, effectiveness, autonomy, and social justice) providing further support for a public health ethics analysis of perceptions of consent for NBS.
Chapter 3. Literature Review

Chapter Overview

This public health ethics framework set the parameters for my review of the extant empirical literature regarding parent and HCP perspectives on consent for NBS in various jurisdictions around the world. The vast majority of this scholarship does not outwardly declare a public health ethics lens of analysis. However, I argue that the principles of public health ethics that I articulated in Chapter 2 are evident in the existing literature (perhaps not surprisingly given the public health nature of NBS), thereby offering further support for my decision to use a public health ethics lens for this inquiry. Using a public health ethics lens in my review of the literature I identified a number of public health ethics themes including expressed tensions between promoting parental autonomy and ensuring high NBS uptake; questions about the perceived effectiveness of informed consent – both implied and express – in achieving its goals of enabling informed autonomous decision-making and protecting individual liberties; and associated implications for social justice upon recognizing that the autonomy of some parents of a given community may be more compromised than others when it comes to express consent for NBS and/or NBS-related research and what that might mean for infants, parents, and society at large. In this chapter I will explore these themes in greater detail.

This chapter will also provide insight into the perspectives of parents and the preferences and needs they articulate in relation to the various dimensions of NBS programs. In addition, I will present the research studies that capture HCPs’ perceptions of the policy
and practice of consent for NBS. Collectively these studies will present the background context within which to situate the consent perspectives of my participants who serve in advisory capacities to the government or as advisors to the advisors.

The Need for Knowledge

A growing number of studies have added an empirical contribution to the ongoing debate regarding consent for NBS. Researchers in Canada, the United States, the Netherlands, the United Kingdom, and Australasia have conducted independent studies that explore parent perspectives on the issue of consent for NBS in their respective jurisdictions. While informing parents and obtaining express consent from parents are two distinct undertakings, parent education and knowledge dissemination arguably lie at the heart of both activities. NBS programs in many jurisdictions have expanded the number of conditions for which infants are screened, store the blood spots anywhere from 2 years to indefinitely (Avard et al., 2006), and use the dried blood spots for a range of purposes, among them research (Bombard et al., 2012). Parents involved in research studies that focus on their experiences with NBS programs and their perceptions of consent for NBS (Faden et al., 1982; Holtzman et al., 1983; Campbell & Ross, 2003; Moody & Choudhry, 2011) as well as studies that engage parents and citizens alike in research addressing the storage and research dimensions of some NBS programs (Tarini et al., 2010; Rothwell et al., 2010; Bombard et al., 2012; Araia et al., 2012) emphasize the importance of parents being informed about NBS, storage, and research.

A study conducted by researchers in Maryland, one of the few states in the United States that has legislated a voluntary NBS program requiring express written parental consent
for NBS, found that most mothers privileged the information about NBS above the presence or absence of an actual consent process (Faden et al., 1982; Holtzman et al., 1983). This finding has since been corroborated over the years by other researchers underscoring that while parents stressed the importance of parents being informed about NBS, parents were often divided as to whether express parental consent for NBS was necessary (Campbell & Ross, 2003; Detmar et al., 2007): “Most parents were less concerned about whether consent was required but more concerned that they were informed” (Campbell & Ross, 2003: 212).

In an Ontario-based survey study involving mothers whose infants had screened negative for the conditions on the newborn screen, researchers found that mothers with higher NBS knowledge scores reported higher overall satisfaction with the program than those mothers with lower knowledge scores (Araia et al., 2012). Holtzman et al. (1983) reported that mothers’ knowledge about NBS influenced their attitudes towards the perceived necessity of consent: mothers in this study found to have “lower knowledge scores” were more likely to want a consent-based model for NBS than women who had a better understanding of the NBS program. These findings arguably reinforce the importance of ensuring, to the extent possible, that parents are well informed about NBS.

Despite parents’ desire for being informed about NBS, researchers in North America, the United Kingdom, and Australia found that in their studies most parents were largely unaware of NBS in general and the screening process, the conditions on the panel, and the treatment possibilities in particular (Campbell & Ross, 2003; Suriadi et al., 2004; Hargreaves et al., 2005b; Grob 2006; Detmar et al., 2007; Parsons et al., 2007; Lang et al., 2009; Moody & Choudhry, 2011). Of the parents who reported being aware of NBS, studies have found
significant gaps in knowledge and understanding, including information about what a false positive screen means (Sorenson et al., 1984; Tluczek et al., 1992; Waisbren et al., 2003; Araia et al., 2012). Even in jurisdictions that purportedly encourage informed decision-making for NBS through express or implied consent processes, studies involving parents have found that some parents who “consented” to NBS for their infants remained uncertain as to what NBS was, what it was they gave their permission for, and/or did not realize that they could have declined screening on behalf of their infants (Faden et al., 1982; Holtzman et al., 1983; Lester et al., 1992; Campbell & Ross, 2003; Hargreaves et al., 2005b; Suriadi et al., 2004; Parsons et al., 2007; Detmar et al., 2007; Moody & Choudhry, 2011; Araia et al., 2012). The findings of these research studies arguably raise questions about the effectiveness of consent, as currently implemented in various jurisdictions, in achieving informed autonomous decisions. The apparent lack of knowledge and understanding, both essential components of informed consent – implied or express – suggests that the practice of consent as currently conducted in many jurisdictions is arguably failing to meet not only the standards of consent, but also the expectations of parents as the surrogate decision-makers for their children that they be informed about the medical interventions that their infants undergo.

Explanations put forth to explain the relative lack of parental knowledge and understanding regarding NBS include HCPs not talking to parents about NBS for reasons of time, cost, or feeling ill-equipped to talk to parents about NBS (Lang et al., 2009; Hayeems et al., 2009; Ross, 2010), and parents themselves not understanding due to sociodemographic influences on knowledge and understanding (e.g., age, marital status, education, insurance,
race and ethnicity, English as a second language, and family history of genetic illness) (Holtzman et al., 1983; Suriadi et al., 2004; Davis et al., 2006; Lang et al., 2009), as well as arguments that recall measures typically used in research studies to assess parental knowledge and understanding might fall short of capturing parents’ true comprehension (Nicholls, 2010).

Other possible factors interfering with parental comprehension can also be attributed to the high reading levels of the NBS information pamphlets. For instance, Faden et al. (1982) found that Maryland’s NBS disclosure form was at a tenth-grade reading level. More recent studies, geared specifically towards evaluating NBS information pamphlets, found that NBS reading material was difficult to understand and incomplete, privileging the “public health agenda” over informed choice (Hargreaves et al., 2005a: 111) and parent understanding (Arnold et al., 2006). Arnold et al. (2006) found that most NBS information materials average between tenth- and twelfth-grade reading level difficulty, despite an average eighth-grade reading level ability among adults in the United States. Patient education and health communication experts suggest that such information brochures be written in a user-friendly manner significantly below an eighth-grade reading level: “Evidence suggests that almost all patients, not only those with limited literacy skills, prefer easy-to-read materials over more-complex or more-comprehensive materials” (Arnold et al., 2006: s321). Researchers engaged in evaluating NBS information and consent materials emphasize the importance of ensuring that the information given to parents is brief, accurate, and written in clear and simple language so that the information about NBS is manageable.
and not overwhelming (Stewart et al., 2005; Hargreaves et al., 2005b; Arnold et al., 2006: s324; Moody & Choudhry, 2011).

A couple of studies examining parent information needs corroborate this recommendation by highlighting parents’ simultaneous desire for more information with less details (Davis et al., 2006; Moody & Choudhry, 2011). Parents in Davis et al.’s (2006) study are quoted as saying,

‘I don’t want a lot of details. I just want it as short and as simple as possible’ and ‘Put less information [in the brochure] so people will read it. Make it more concise, less overwhelming.’ (p.s332)

Similarly, Hargreaves et al. (2005b) found that most parents, “with hindsight” saw “no need for detailed information about the screening tests or the conditions tested for before, or at the time of, the test” (p.165). That said, parents with affected children, as well as parents who could imagine receiving a positive screen, expressed wanting to know more about NBS and the conditions on the panel prior to the heel prick (Hargreaves et al., 2005b). A study conducted by Moody and Choudhry (2011) highlighted that ultimately parents have different information needs (e.g., some wanting more information and others wanting less) and that “[p]arents should be empowered to choose information relevant to their own decision-making approach and information processing capabilities” (p. 9).

These findings collectively show that parents may be either uninformed or under-informed about NBS, which suggests that many parents are likely underserved by information efforts and informed consent processes designed and intended to respect parents as the surrogate decision-makers for their children. Coupled with the sociodemographic influences on knowledge and understanding within the context of informed consent (implied
or express), these findings also underscore the social justice implications of both NBS information dissemination and consent processes and, as a result, the need for social justice considerations when examining consent policy and practice. As articulated in Chapter 2, and reiterated here in the empirical findings of existing scholarship, social determinants of health can have implications not only for health, but also personal security, reasoning capacity, self-respect and the respect of others, attachment, and the ability to exercise autonomy through self-determination (Powers & Faden, 2006). As a result, approaches to information dissemination and consent regarding NBS arguably need to consider the range of parental information needs reflected in the diversity of a given community.

**Tensions Between Respecting Parental Autonomy and Achieving High Uptake**

Although parents in these studies expressed wanting to be informed about NBS, many researchers reported that parents were divided on the issue of express, written, parental consent. Studies found that parents wrestled with the possibility of express consent for NBS, appreciating on the one hand the perceived challenges that such an approach to consent could potentially have for the uptake of NBS and the associated implications for infant health, but also recognizing on the other hand that parents are ultimately the surrogate decision-makers for their children and that the decision about whether their infant should undergo NBS should be theirs to make (Campbell & Ross, 2003; Hargreaves *et al.*, 2005b; Quinlivan & Suriadi, 2006; Moody & Choudhry, 2011).

Some parents in these studies described their perceptions of NBS as low risk (Moody & Choudhry, 2011), routine (Parsons *et al.*, 2007), beneficial, “nationally recommended” (Quinlivan & Suriadi, 2006: 69), and necessary to prevent infant harm —
ultimately a program in the “interests of their children’s health” (Detmar et al., 2007: 242). Parents of children diagnosed with one of the conditions on the NBS panel were among the parents most likely to endorse “routine” screening (Hargreaves et al., 2005b: 164). In three independent studies parents raised the concern that express consent introduced a risk that “other parents” might not be trusted to make “the right” decisions “in the best interest of their child” (Campbell & Ross, 2003: 210; Detmar et al., 2007: 241; Moody & Choudhry, 2011: 8). Some parents in these studies expressed particular concern that parents – young, teenage parents in particular – might decline NBS on behalf of their infants purely out of ignorance (Campbell & Ross, 2003). Similarly, Detmar et al. (2007) reported in their study that parents expressed concern for those parents whom they “suspected would not be able to cope with the freedom of choice” (p.241).

Other parents, however, stressed the importance of parental consent for NBS (Hargreaves et al., 2005b; Quinlivan & Suriadi, 2006). Some parents supported their position by identifying parents as the rightful surrogate decision-makers for their children (Campbell & Ross, 2003; Moody & Choudhry, 2011). Others situated their preference for parental consent on their desire to know everything that happens to their baby (Faden et al., 1982). Some parents endorsed express consent because they wanted to be able to decline NBS for reasons including religious beliefs and due to their perceptions that NBS was either not beneficial or not essential for their child’s health (Campbell & Ross, 2003; Quinlivan & Suriadi, 2006). Although many parents in these studies indicated a strong preference for an informed consent approach, express written consent was not necessarily needed; rather, provided HCPs presented parents with a clear choice and the information necessary to make
that choice, the majority of parents in Moody and Choudhry’s (2011) study endorsed an opt-out approach to consent:

In terms of consent, a system of informed dissent or opting out, in line with current service provision was seen to be desirable by nearly three quarters of participants. However, improvements could be made to existing informed dissent/choice model with the optional nature of screening being made more transparent. Whilst a formal signature from the parent to document consent may lead to reassurance that information had been adequately relayed and a decision consciously made, it was not deemed necessary by the majority of participants. (p. 10)

However, while parents in many of these studies conveyed their preference for explicit choice for NBS, they articulated challenges that they perceived would accompany such a decision. For example, some parents who supported informed consent for NBS – wanting the power to make decisions on behalf of their children – also displayed a sense of inner conflict introduced by the perceived gravity of the decision and burden of information (Detmar et al., 2007; Moody & Choudhry, 2011). In fact, some parents revealed that while they advocated for an informed choice-based approach to NBS, they were ultimately glad that when their infants were screened that they did not have to make a choice (Detmar et al., 2007). Some parents identified the source of their stress in the realization that in order to make an informed decision they would need more information, but they felt they would be ill-equipped to “appraise the information” (Detmar et al., 2007: 242). Detmar et al. (2007) underscored the challenge of providing parents with the information necessary to facilitate informed decision-making without sending the parents into information overload. In Moody and Choudhry’s study (2011), some parents described their perception of express, written consent as a warning sign that would “arouse suspicion and mistrust, causing extra worry and making the decision harder” (p.8). Finally, some parents who advocated for parental consent
identified the tension that honouring parental autonomy would likely mean that some parents might make “‘a reckless decision that’s going to impact on their children’s health,’” but that ultimately “‘they’ve probably got the right to take it, but I’m really not sure...it’s a very difficult one’ [CF father]” (Hargreaves et al., 2005b: 166).

Similarly, attitudes towards consent for NBS from the perspectives of HCPs charged with informing parents about NBS and/or obtaining parental consent, reveal similar tensions between HCPs wanting to respect parental autonomy by ensuring parental consent while also having mandatory screening programs to ensure program uptake and promote infant health (Huang et al., 2005; Kerruish et al., 2008; Hayeems et al., 2009; Miller et al., 2010b). In a New Zealand study conducted by Kerruish et al. (2008) exploring the attitudes of lead maternity carers towards informed consent for NBS, these researchers found their participants described contradictory preferences, wanting NBS programs to be both voluntary and mandatory:

Of those who considered that NBS should be mandatory, most (89%) still believed, somewhat paradoxically, that some form of parental consent should be obtained. Moreover, of those who thought testing should not be mandatory, only a small proportion (10%) would accept parental refusal without question. (p. 651)

Kerruish et al. (2008) interpret these findings as illustrative of “the complexity involved when the public health aim of maximising the uptake of testing coexists with a commitment to respect parental autonomy” (p. 650). In an Ontario-based mixed methods study conducted by Miller et al. (2010b), among the outcomes reported from this research include the finding that while the approach to consent for NBS in Ontario is implied, many nurses and midwives purport to obtain express verbal or written consent:
A large majority of midwives (92.2%) and a substantial minority of nurses (39.2%) reported obtaining verbal or written consent (defined as consistently or usually) to NBS; by contrast, only tiny minorities of physician respondents reported doing so (3.6%-4.5%). Consent practices were closely aligned with the practical task of physically taking the blood sample from the infant. (p. 184)

Conversely, in Taiwan, hospitals and clinics have implemented a tiered approach to consent for NBS with a mandatory core panel of five conditions and two additional screening options requiring express consent (Huang et al., 2005). However, Huang et al. (2005) found that in many clinics and hospitals the supplemental screening was routinely conducted without seeking parental consent:

In this study, over half the hospitals/clinics studied did not adhere to an “informed consent” model for screening rare metabolic/genetic disorders, and the information provision rarely included the treatment limitations. Hence, the way most NBS is currently conducted is in conflict with basic ethical principles. (p. 624)

Explanations put forth for this disconnect reiterate the tension between respecting parental autonomy and fulfilling the goals of NBS, namely “to protect newborns’ health” (Huang et al., 2005: 624). These scholars conclude that at the very least, regardless of whether NBS is mandatory or voluntary, parents need to be informed:

Whether or not any category of NBS should be voluntary or mandatory, informing parents before screening is necessary to respect the parental right to know and protect parental autonomy. (Huang et al., 2005: 624)

Collectively these research studies that have explored the attitudes and preferences of parents and HCPs regarding consent for NBS underscore the perceived challenge of balancing the desire to achieve public health goals while also respecting parental autonomy. The sustained refrain across these studies, however, emphasizes that independent from the approach to consent, parents need to be fully informed about NBS. Among the perceived challenges associated with this undertaking – as documented in the literature – pertains to
when such information dissemination should occur. In the section below I provide a synthesis of parent preferences on this issue.

**When to Inform and/or Obtain Consent: Parents’ Perspectives**

In light of the perceived volume and complexity of the information that parents felt would accompany an informed consent process for NBS, not to mention the perceived seriousness and gravity of the decision, parents were adamant that they did not want to be informed about NBS postpartum (Davis *et al*., 2006; Detmar *et al*., 2007; Parsons *et al*., 2007; Moody & Choudhry, 2011). Parents described their time in the hospital as “a fog” and insisted they were in no condition to learn about NBS for the first time following the birth of their child (Davis *et al*., 2006). Studies have found, however, that in some jurisdictions such as Ontario, postpartum is often exactly when parents learn of NBS:

Among respondents who reported having received information about NBS (n=520), a majority recalled receiving this information after the birth of the infant, either just before the NBS test (62%) and/or at the time of the heel prick test (72%). (Araia *et al*., 2012: 965)

Parents expressed a preference to learn about NBS at some point during pregnancy instead (Detmar *et al*., 2007; Parsons *et al*., 2007; Moody & Choudhry, 2011). Some parents advocated for continuous NBS education beginning mid-pregnancy, following labour and delivery, and again right before the heel prick (Moody & Choudhry, 2011) and others expressed wanting to learn about NBS pre-conception (Detmar *et al*., 2007). Finally, in addition to wanting to know about NBS, studies have found that parents also want to be informed about the storage and research dimensions of NBS as well — the focus of the next and final section of this chapter.
Parental Attitudes and Responses to Dried Blood Spot Storage and Research

A primary argument put forth against express consent for NBS is the fear that if parents were told, for example, that the dried newborn blood spots are stored or that different kinds of research may be conducted on the blood spots, the more likely parents would be to decline NBS altogether, in so doing leaving their infants vulnerable (Avard et al., 2006; Rothwell et al., 2010; Richer et al., 2011). Studies exploring parent perceptions of, receptiveness towards, and participation in these expanding dimensions of NBS suggest that parents are perhaps more open to these possibilities than initially assumed (Stolt et al., 2002; Campbell & Ross, 2003; Lernmark et al., 2004; Feuchtbaum et al., 2006; Feuchtbaum et al., 2007; Skinner et al., 2011).

A number of studies have ascertained parent perceptions of consent for the storage of and future uses for blood spots including research (Tarini et al., 2010; Rothwell et al., 2010; Bombard et al., 2012; Rothwell et al., 2012; Botkin et al., 2012). The growing consensus within the literature suggests that parents want to be informed about and have an opportunity to consent to the storage of and research on dried blood spots (Quinlivan & Suriadi, 2006; Tarini et al., 2010; Rothwell et al., 2010; Rothwell et al., 2012; Bombard et al., 2012). In terms of parental willingness to participate in such initiatives, Tarini et al. (2010) found that provided parents had an opportunity to consent, three quarters of the participants reported that they would be “very or somewhat willing” to have their infants’ blood spots used for research purposes. Without express consent, however, those numbers dropped significantly to 28.2%, suggesting that public health research initiatives can be achieved by cultivating trust through transparent processes (Tarini et al., 2010; see also Bombard et al., 2012). With
respect to storage of dried blood spots, 78% of parents in Tarini et al.’s study (2010) would allow their infants’ blood spots to be stored, whereas participants in Rothwell et al.’s study (2010) were clear that express consent for the storage of blood spots should be sought prior to storage, even if the blood spots would be stored anonymously. Most participants in this study expressed wanting the samples destroyed immediately following the screening process (Rothwell et al., 2010). Participants in Bombard et al.’s study (2012) supported blood spot storage for infant health and quality control purposes as well as for future anonymous research, yet participants were divided as to whether express consent should be sought for future research. In Bombard et al.’s study the apparent tensions between individual and population interests framed the challenge of how to proceed with consent:

Our value elicitation exercise does not provide clear direction regarding parental choice, however, in part because the balance between individual and collective interests is difficult to establish, and because this balance concerns a research enterprise that is situated within a public health program. (p. 245)

A number of studies have examined actual parent participation in NBS-related research and reported on the experiences. In studies where parents were approached to participate in NBS-related research projects such as consenting to have their infant screened for a supplementary panel of conditions (Feuchtbaum et al., 2007), or Fragile X syndrome (Skinner et al., 2011) or Type 1 diabetes (Stolt et al., 2002; Stolt et al., 2003; Lernmark et al., 2004), researchers reported that parents were appreciative of the opportunity to participate in the study (Feuchtbaum et al., 2007); they perceived themselves as the appropriate surrogate decision-makers for their children in such contexts (Stolt et al., 2002); and they were, for the most part, willing to participate (Stolt et al., 2002; Lernmark et al., 2004; Feuchtbaum et al., 2006; Feuchtbaum et al., 2007; Skinner et al., 2011). Feuchtbaum et al. (2007) and Stolt et
al. (2003) found that many parents wanted their infants screened for conditions, even if nothing could be done in the event of a positive diagnosis. Many parents felt it was their right to learn about any incidental findings generated through NBS, since such findings pertain directly to their infant’s health (Stolt et al., 2002).

These studies suggest that parents are perhaps more open to the expanding purview of NBS-related initiatives (e.g., storage, supplemental screening research, and/or other research endeavours) than initially thought, provided informed parental consent is sought. As evident in Rothwell et al.’s (2010) study, parents expressed wanting to be informed about and have a choice in such matters as blood spot storage and post-screen uses of the blood spots following the newborn screen. Of course, wanting to be informed and to have a choice about future uses of newborn blood spots does not mean that all parents will ultimately consent to such uses (Rothwell et al., 2010). However, as evidenced in the literature presented above, one cannot necessarily conclude that parents who refuse the storage and research dimensions of NBS programs will ultimately decline the initial screen as well.

**Chapter Conclusion**

In conclusion, this chapter has shown that research is beginning to demonstrate empirically that some of the worst fears harboured by critics of express consent for NBS — namely that parents would decline screening on behalf of their infants if they are required to give express consent for NBS, blood spot storage, and research — may be more theoretical than real. However, existing research on current approaches to informed consent for NBS – both express and implied – also shows that legitimate questions can be asked about whether the practice of consent succeeds in achieving its goal of promoting individual autonomy
through self-determination and informed decision-making or whether consent is merely a
perfunctory process that simply creates the illusion of choice and respect for persons.
Connected to these challenges of protecting and promoting self-determination through
consent processes are the sociodemographic variables found to influence knowledge and
understanding and ultimately choice within such contexts. Perhaps most importantly, these
studies show that independent from consent, parents want to be informed about NBS, the
storage of dried blood spots, and the possibility of future uses for dried blood spots such as
research. Finally, the sustained characterization of tensions in debates around consent for
NBS, storage, and research as being between the individual and collective good, or between
parental autonomy and NBS uptake, or between parental autonomy and infant health
provides additional support for the appropriateness of a public health ethics lens of analysis
for consent as a least restrictive alternative for NBS in Ontario. These findings provide a
foundation for my public health ethics inquiry of consent as a least restrictive alternative to
achieve NBS health goals that draws on the public health ethics principles of autonomy,
effectiveness, and social justice.
Chapter 4. Methods

Chapter Overview

I conducted an exploratory, qualitative case study to examine ethical issues related to NBS in Ontario with a particular focus on the issue of consent. In this chapter I provide a general introduction to case study research followed by a description of my case study inquiry. The remainder of the chapter outlines the multiple sources of data drawn upon for this research; details the sampling and recruitment strategies used to recruit participants; describes the research settings and interview process; explains the data analysis and interpretation; articulates efforts to ensure rigour and trustworthiness; and explains ethical challenges that emerged during this study.

Introduction to Case Study Research

Case study research is an approach to qualitative and/or quantitative inquiry typically used to answer how, why, and, in the case of some exploratory studies, what research questions (Yin, 2003). Case study research can take many forms and can assume a variety of labels: a single or multiple case study (Yin 2003; Stake 2005), an intrinsic or instrumental case study (Stake, 2005), or an explanatory, descriptive, or exploratory case study (Yin, 2003). Ultimately, case study research seeks to understand complex phenomena that are deeply situated and engaged in the physical, social, political, economic, cultural, ethical, and historical contexts of its environment (Yin, 2003; Stake, 2005; Baxter & Jack, 2008). The comprehensive investigation, deconstruction, and reconstruction of a case can lead to
generalizations that extend beyond the phenomena to address more abstract or theoretical concepts (Yin, 2003; Stake, 2005).

Different case study research philosophies exist. At the most fundamental level, scholars disagree as to whether case study research is a methodology. Robert Yin (2003), for example, considers a case study “an all-encompassing method—covering the logic of design, data collection techniques, and specific approaches to data analysis … a comprehensive research strategy” (p.14), although he concedes that research on case study methodology to date has not articulated a well-defined approach for the interpretation of findings. Conversely, Robert Stake (2005) insists that case study research “is not a methodological choice but a choice of what is to be studied” (p. 443) – a position shared by a number of other case study research scholars as well (Creswell, 2007; Merriam, 2009). These scholars argue that a number of qualitative and/or quantitative research methods can be used effectively to study a given case (Merriam, 1998; Stake, 2005; Creswell, 2007; Merriam, 2009). Merriam (1998), who writes about qualitative case study research in particular, is clear that approaches to qualitative case study analysis can take many different forms including “basic or generic qualitative research, ethnography, phenomenology, or grounded theory” (p.12).

Other differences in approach pertain to the use of theory in case study research. Stake (2005) contends that theory should only be integrated towards the end of the project given that “[o]ne cannot know at the outset what issues, perceptions, or theory will be useful” (p. 456). Yin (2003), on the other hand, stresses the importance of integrating theory from the outset regardless of whether the goal of the study “is to develop or test theory” (p.
Such theoretical assertions, Yin (2003) argues, serve to guide research design and analysis despite the fact that new issues or ideas might emerge, thus requiring further theoretical consideration and/or an adjustment to ensure the theory captures, in its evolution, the most relevant dimensions of the case.

Although scholars differ in some aspects of their respective approaches to case study research, they agree on the importance of selecting a structured case with definitive boundaries and, whenever possible, incorporate multiple sources of data (Yin, 2003; Stake, 2005; Creswell, 2007; Merriam, 2009). First, establishing the case – or “bounded system” – to be studied is among the most challenging dimensions of a qualitative case study and often the most confusing (Creswell, 2007; Baxter & Jack, 2008; Merriam, 2009). A case can, for example, focus on an individual, a group of individuals, a program, an intervention, a policy, an event, an institution, a community, an issue, or some other social phenomenon (Merriam, 1998; Hancock & Algozzine, 2006; Baxter & Jack, 2008; Merriam, 2009). Baxter and Jack (2008) underscore that in qualitative case study research the phenomenon under examination is often inextricably bound to the context, in that “the case could not be considered without the context” (p.545) (e.g., ethical issue = case, NBS in Ontario = context).18

Once researchers have identified their case, they must set parameters on the case – or “bind” the case – in an effort to make sure that the inquiry is manageable (Yin, 2003; Stake, 2005; Creswell, 2006; Baxter & Jack, 2008; Merriam, 2009). Qualitative case study researchers might consider binding their case according to time, place, events, processes, or activities (Hancock & Algozzine, 2006; Creswell, 2007; Baxter & Jack, 2008). Although

18 See also Hancock and Algozzine (2006) and Merriam (2009) for more information about the interconnection between a case and its context.
these scholars emphasize the importance of placing appropriate and sufficient parameters on the case, they recognize that the constructed boundaries may shift necessarily once the research is underway (Yin, 2003; Stake, 2005; Merriam, 2009): “Ideally, for example, the design of a qualitative study is emergent and flexible, responsive to changing conditions of the study in progress” (Merriam, 1998: 8).

Second, case study researchers often, but not always, incorporate multiple data sources to illuminate the case under inquiry (Yin, 2003; Creswell, 2007; Baxter & Jack, 2008; Merriam, 2009). Data sources frequently used in qualitative case study research include, for example, interviews, focus groups, observations, documents, archival records, and artifacts (Yin, 2003; Stake, 2005; Creswell, 2007; Baxter & Jack, 2008). In some case studies one source of data will dominate (e.g., interviews, documents, or observation) and researches will supplement that data by drawing on additional sources (Merriam, 2009). Including multiple sources of data in an inquiry allows researchers to acquire additional insights; to challenge and/or support existing data; to generate inferences; and to establish or verify case “facts” (Yin, 2003; Creswell, 2007; Baxter & Jack, 2008). These data are then interwoven throughout the case study narrative to provide a more comprehensive understanding of the case (Baxter & Jack, 2008).

In short, qualitative case study research is used “to gain an in-depth understanding of the situation and meaning for those involved” (Merriam, 1998: 19). The goal of qualitative case study research is not to test hypotheses, but rather to make discoveries, gain fresh insights, offer new interpretations, and/or in some instances, support what has already been established:
‘Previously unknown relationships and variables can be expected to emerge from case studies leading to a rethinking of the phenomenon being studied. Insights into how things get to be the way they are can be expected to result from case studies.’ (Stake, 1981, p.47 cited in Merriam, 2009: 44)

The final “write-up” of qualitative case study research is necessarily more descriptive than other qualitative works given that researchers need “to convey a holistic understanding of the case” (Merriam, 2009: 204; see also Hancock & Algozzine, 2006). However, qualitative case study research often extends beyond description to include unique substantive and/or theoretical insights and interpretations that can contribute to the development and/or evaluation of new programs, interventions, policies, and practices; the advancement theory; and/or the introduction of new questions to be asked in future research inquiries (Creswell, 2007; Baxter & Jack, 2008; Merriam, 2009).

**My Exploratory Qualitative Case Study**

Given the diverse and often nuanced interpretations and applications of case study research I have drawn predominantly on Merriam’s qualitative approach to case study inquiry (1998; 2009). She emphasizes the importance of identifying and setting parameters on the case, whether it be a person, a program, a policy, or issue, yet argues that any number of qualitative approaches to data collection, analysis, and interpretation may be used in a qualitative case study (Merriam, 1998; 2009). I designed a single, exploratory, qualitative case study to explore ethical issues related to NBS in Ontario. In addition to placing boundaries on my case according to place (Ontario), I also limited my inquiry by time and event: specifically, beginning November 2005, when the Ontario Ministry of Health and
Long-Term Care announced the expansion of Ontario’s NBS program, to the present.\textsuperscript{19} Interview data with individuals involved in some capacity with the design, implementation, and/or advocacy of expanded NBS in Ontario serve as my primary source of data supplemented by observation and document data. The exploratory dimension of my study is captured in the wide cast of my initial inquiry of ethical issues. I wanted to know what my participants identified as ethical issues related to NBS in Ontario both at the time of the expansion and at the time of our interview (which occurred between February 2010 and January 2011). Within the context of the interviews, participants discussed many ethical issues, among them parental awareness; parental and HCP education; consent; carrier status disclosure; false-positives; and storage of and research on dried newborn blood spots. Each of these ethical issues could have been the focus of an independent case study. However, through the interview and analysis processes (explained in greater detail below) I decided to focus on the policy and practice of consent given that many of the ethical issues introduced by my participants converged either directly or indirectly with the issue of consent for NBS in Ontario.

As a result, this qualitative case study explores how key informants\textsuperscript{20} charged with advising the Ontario Ministry of Health and Long-Term Care, either directly through their

\textsuperscript{19} Although my interviews occurred between February 2010 and January 2011, I continued to track relevant documents through May 2013.

\textsuperscript{20} I draw on Thorne’s (2008) definition of the term “informant” to justify my use of the label to describe my participants: “The term ‘informant’ has a long history within ethnographic circles, and reflects those individuals within a culture or society identified as being particularly familiar with the relevant elements of the culture and who were willing to spend the time to explain them to you” (Thorne, 2008: 93). In addition, Merriam (1998) underscores that “informants” not only understand the culture or society, but “are also able to reflect on it and articulate for the researcher what is going on” (p.85). Within the NBS culture in Ontario, my participants or “key informants” were involved in some capacity with the expansion of Ontario’s NBS and, therefore, would be able to shed light on what they perceived to be among the most salient ethical issues connected to the program from their unique vantage point.
position as a member of one of the past or present provincial NBS advisory committees or indirectly through consultation (e.g., HCPs working at the NBS treatment centres) and advocacy (e.g., individuals who championed expanded NBS in Ontario), perceive the policy and practice of consent for NBS in Ontario. This inquiry seeks to examine and interpret the intellectual substance and significance my participants attribute to the phenomenon of consent and to explore the broader theoretical and social questions of consent in a public health context.

Sources of Data Used for this Inquiry: Interviews, Observation, & Documents

One-on-one interviews comprise the primary source of data for this exploratory qualitative case study. Additionally, I supplemented this data with observation field notes and documents. These data are integrated throughout my dissertation (Baxter & Jack, 2008) to help frame the case, provide case “facts,” as well as complement and challenge data and arguments. Here I provide a full list of my sources of data.

Interviews

I conducted 57 one-on-one interviews between February 2010 and January 2011. Interviews were transcribed and transformed into approximately 1400 pages of data, which serve as the main source of data for this inquiry. Given the primacy of these interviews, following the overview of the data sources used in this study the remainder of the chapter will be devoted to describing my sampling and recruitment strategies, research setting, and interviews followed by a comprehensive description of my analysis and interpretation.
Observation Field Notes

I attended and observed Newborn Screening Ontario’s Clinical Follow-Up Symposium on April 16, 2010. Symposium attendees included members of the Newborn Screening Ontario Team as well as genetic counsellors, nurses, and physicians who work at the five NBS treatment centres across the province. I have drawn on my comprehensive field notes from observing a round table discussion to highlight the lengths Newborn Screening Ontario and the treatment centres go to in order to ensure, to the extent possible, that all infants who screen positive for a condition on the screening panel receive the necessary follow-up testing.

Document Data

In my qualitative case study inquiry I have incorporated a number of documents ranging from brochures and blood spot requisition forms, to newspapers and websites, to policies and legislation.

Brochure

I have drawn on Ontario’s Newborn Screening brochure entitled, “Newborn Screening: A healthy start leads to a healthier life” (See Appendix F)\(^{21}\) to show that NBS in Ontario is currently framed to parents as something that will happen to their infants rather than as a choice parents have to make. However, the brochure does provide the website address for parents who want more information.

\(^{21}\) This was the brochure that was in circulation when I submitted my research proposal to ethics in the Fall of 2009.
Parent information sheet

In Ontario, after the blood spots have been taken from the infants’ heels, parents are meant to receive a Parent Information Sheet alerting them to this fact. This parent information letter is a “tear-off” sheet attached to the blood spot requisition card that serves as an informational “receipt” (as the serial number on the top right-hand corner matches the blood spot card sent to the screening lab should parents wish to follow-up). This letter informs parents that blood was taken from their infant’s heel for NBS; very briefly explains what the screen is for (for parents who may not know); and discloses that “[p]ersonal health information will be shared between the health care providers involved in newborn screening and diagnosis to ensure your baby receives appropriate care and follow-up” (see Appendix G). Parents with additional questions are directed to contact their HCP. I have incorporated this document to show that parents are not directly informed about the storage and research dimensions of NBS in any of the written materials they are supposed to receive.

Report by the Provincial Advisory Committee on New Predictive Technologies

I drew on the report written by the Provincial Advisory Committee on New Predictive Technologies entitled, Genetic Services in Ontario: Mapping the Future, which was published online in 2001. This report offers a definition of what constitutes informed consent in Ontario. In addition, regarding NBS in Ontario specifically, the committee recommended – prior to the program’s expansion – that express, written consent should be required: not only for the newborn screen, but also for the storage of NBS data and samples. The

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22 Although parents are supposed to be given this information sheet, the Newborn Screening Ontario Lab purportedly receives a lot of blood spot cards with the parent letter still attached (treatment centre, #21).

23 The parent information sheet I reference was the form in circulation in 2009.
committee also suggested that the government assemble an appointed body to examine whether and when NBS should be permitted in the absence of express parental consent.

**Terms of reference for the provincial newborn screening advisory committees**

I also drew on the terms of reference documents for the NBS advisory committee that was established at the time of the program’s expansion (Ontario Advisory Committee on Newborn and Childhood Screening (Ministry of Health and Long-Term Care, 2006)) as well as the current advisory committees: the New Provincial Maternal-Child Screening Committee (Born Ontario, 2013a) and Prenatal and Newborn-Child Sub-Committee (Born Ontario, 2013b). These documents outline the terms of reference for the advisory committees including the committee mandates and activities. I have used these documents to demonstrate that the information available to the public about the first committee’s mandate did not make reference to a focus on ethical issues related to NBS (among my reasons for setting my case study parameters on issues of ethics). Since then, ethical issues connected to NBS in Ontario, among them consent, have been publicly identified in the committee’s mandate.

**Ombudsman report**

For this case study, the Ombudsman’s report published in 2005 entitled, *The Right to be Impatient*, provided background information about the expansion of Ontario’s NBS program (Marin, 2005).
News Media

To defend my framing of NBS as a public health program in Ontario I incorporate an excerpt from an online newspaper article by Honeywell and Cloutier (2007) that was featured in *HNews Canada’s Health-Care Newspaper* in 2007 following the program’s expansion.

Websites

Newborn Screening Ontario’s website (newbornscreening.on.ca) served as an additional source of data. I used this website as a resource to establish details about the past, present, and future of NBS in Ontario including the conditions for which the province screens (see Appendix B); the steps necessary to obtain carrier status results for sickle cell anaemia; and information about the storage of samples, reasons behind storing the sample, possible future research uses of samples, and the steps parents need to take to have their infants’ samples destroyed should they so choose. I also included an excerpt from the FAQ section regarding whether parents may refuse screening to challenge the “voluntariness” of consent for NBS in Ontario (see Newborn Screening Ontario, n.d.g).

Codes, policies, and legislation on consent

To establish the definitions of and criteria for informed consent in health care I turned to seminal international and provincial documents. First, the Nuremberg Code (1947) is a document that stemmed from the Nuremberg Trials at the end of World War II. The need for “voluntary consent” is the first of 10 research ethics principles outlined in this international code. The regional policies and legislation I used include the *Consent to Medical Treatment Policy* put forth by the College of Physicians and Surgeons of Ontario in 2006 and Ontario’s *Health Care Consent Act 1996*, respectively.
Published articles

Yin (2003) notes that “[f]ormal studies or evaluations of the same “site” under study” (p.86) are among the useful documents that can be used to support or challenge data, confirm “specific details” of one’s inquiry, and/or to generate inferences. To demonstrate that the language of public health is still used to describe NBS in Ontario I referred to the following scholars’ work: Miller et al. (2009), Hayeems et al. (2009), Miller et al. (2010), Araia et al. (2012), and Bombard et al. (2012). To confirm that implied consent is the current label used to describe the consent process for NBS in Ontario I drew on the following articles: Hayeems et al. (2009), Miller et al. (2010), and Bombard et al. (2012). Finally, a law review article by Wildeman and Downie (2001) confirmed that implied consent was also the approach to NBS in Ontario prior the program’s expansion as well, not only in Ontario but Canada more broadly.

Having outlined the various data sources that I incorporated in my inquiry, I will now explain in greater detail the approaches I took to sampling, recruiting, and interviewing my study participants.

Participant Sample Strategy and Rationale

Most qualitative researchers draw on non-probability sampling strategies for their studies, the most common of which is “purposive” or “purposeful” sampling (Merriam, 2009). Purposeful sampling is typically used when “the investigator wants to discover, understand, and gain insight and therefore must select a sample from which the most can be learned” (Merriam, 2009: 77). Participants are identified and recruited for their experiential knowledge of and insights into the phenomenon under study (Thorner, 2008; Merriam,
2009). Thorne (2008) underscores the importance of “strategic identification of ‘key informants’” within purposive sampling, stressing that

[t]he rationale for key informants is that some members of a community will be better equipped than others to provide you with access to what is happening and why it is happening. (Thorne, 2008: 91)

Researchers can choose among a number of different types of purposeful sampling methods, among them “typical, unique, maximum variation, convenience, and snowball or chain sampling” (Merriam, 2009: 78). Within the context of qualitative case studies, even though cases should, ideally, be sufficiently bound to generate a limited number of potential participants, Merriam (2009) insists that unless investigators intend to interview all the people within the case, researchers must draw on one of the aforementioned sampling strategies.

For my qualitative case study inquiry I used both maximum variation and snowball sampling strategies. Maximum variation sampling requires researchers to gain insight into the phenomenon under inquiry by identifying and recruiting individuals “who represent the widest possible range of the characteristics of interest for the study” (p.79):

‘Any common patterns that emerge from great variation are of particular interest and value in capturing the core experiences and central, shared dimensions of a setting or phenomenon.’ (Patton, 2002, p.234 cited in Merriam, 2009: 79)

Snowball sampling strategies require that researchers ask their study participants to recommend other individuals “who exemplify the characteristics of interest in the study” (Merriam, 2009: 79).

To this end, I purposively targeted three distinct, yet interconnected, groups of participants, all of whom have arguably been the impetus behind Ontario’s expanded NBS
program: 1) members of any one of the many NBS advisory committees that have operated formally (and informally) in Ontario over the years; 2) HCPs from all five treatment centres across the province; and 3) patient-, parent-, HCP-, and other community-advocates heavily involved in lobbying the government for an expanded NBS program. Each of these groups are comprised of experts with diverse areas of professional expertise and personal
experiences with NBS in Ontario.

I targeted past and present members of the NBS advisory committees because the Ontario Ministry of Health and Long-Term Care explicitly states, in their November 2, 2005 press release announcing the creation of the Ontario Advisory Committee on Newborn and Childhood Screening, that the government will look to this advisory committee for guidance on NBS policies, guidelines, and other emergent issues:

In addition, a permanent advisory committee will be established to provide oversight and ongoing advice to the government on its newborn screening program. (Ministry of Health and Long-Term Care, 2005a)

The committee’s mandate states that among its goals is

[t]o advise the Ministry of Health and Long Term Care on its policies and programs related to newborn and childhood screening. (Ministry of Health and Long-Term Care, 2006)

I interpreted the broad committee mandate to include existing and emergent ethical issues, even though the language of ethics was not explicitly used. Since ethical issues had not been incorporated explicitly into this committee mandate when I began my inquiry, I wanted to understand what the individuals charged with advising the Ontario government on NBS
perceived as ethical issues related to NBS (if any) and what their perceptions of those ethical issues were (particularly given the debates in the academic literature at the time).

Having identified the Ontario Advisory Committee on Newborn and Childhood Screening as the point of departure for my study sample, I turned to the committee membership list to see what other perspectives would be necessary to achieve maximum variation within my study sample. The membership list revealed that nurses and genetic counsellors who work at the NBS treatment centres and who are involved in the care of infants (and their parents) who screen positive for one of the conditions on the newborn screen were not represented among the membership. Given their professional experience working with and caring for screen positive infants and their parents, I thought that including these HCPs in my study sample would provide essential perspectives on existing and emergent ethical issues connected to the daily practice of caring for individuals affected by Ontario’s NBS program.

Advocates comprised the third group of study participants. The significant contribution of patient-, parent-, HCP-, and community advocates in the effort to expand
Ontario’s NBS program had been well documented by the time I was designing my study (Marin, 2005; Eggertson, 2005). Their sustained advocacy for NBS in Ontario necessarily connected them to Newborn Screening Ontario and the Ministry of Health and Long-Term Care. Incorporating the voices of advocates in this study provided the possibility for introducing a different perspective from another group of influential individuals committed to Ontario’s NBS program. These advocates offered another avenue through which to achieve maximum sample variation.

I identified prospective participants for these three specific groups of key informants in two ways. First, I reviewed the grey literature (e.g., newspaper articles, the Ombudsman’s report, and the NBS program websites (both the Ministry of Health and Long-Term Care and Newborn Screening Ontario’s websites)), which revealed a host of potential participants who either were or had been involved as government advisors and advocates. With respect to sampling prospective participants from the 5 treatment centres, I called the telephone numbers for the treatment centres provided on Newborn Screening Ontario’s website. From there I was able to get the names and email addresses of the HCPs actively involved in the care of infants (and their parents) who screen positive for one of the conditions on the NBS panel.

Given the exploratory nature of my inquiry, I wanted to learn from my participants whom they identified as key individuals in the expansion of Ontario’s NBS program and whose voices they thought I should include in my inquiry of perceptions of ethical issues connected to NBS. I did not want to limit my study sample to those individuals whom I perceived to be key players. During the interviews, participants would either spontaneously
reference a name of someone that they felt I should speak with or participants would reveal names at the end of the interview when I asked, “Is there anyone you think I should speak with?” This snowball sampling recruitment strategy generated the names of individuals serving on (or who had once served on) a NBS advisory committee, HCPs in the treatment centres, and advocates. Perhaps most notably, however, this sampling strategy led to the creation of a fourth participant group: legal experts. Since my participants recommended that I include in my sample a handful of lawyers and legal scholars (referred to collectively as legal experts) whom they perceived as having contributed directly or indirectly to discussions around consent for expanded NBS, I did. The decision to include this group in my study and as part of my findings is consistent with the nature of an exploratory inquiry. In addition, including these voices in my study is another way in which I worked to achieve maximum variation.

**Recruitment Strategy & Research Setting**

I primarily contacted prospective participants via email. In the case of advisory committee members and some advocates, their names and contact information were publicly accessible. For prospective participants whose names I received through snowball sampling, participants would sometimes give me the necessary contact information. If they did not have the contact information on hand I was able to acquire it through Internet searches. In the instances where I could not find email addresses to initiate contact, such as with HCPs at the treatment centres and some advocates, I telephoned them first and then if they expressed interest in my study I followed up with an email.
In each unique email I briefly introduced myself, my study, and, based on what I knew about the individual’s involvement (past or present) with NBS in Ontario, why I wanted to interview them. I attached a PDF version of the recruitment invitation to each email (see Appendix H: Letter of Invitation). If I did not receive a response from potential participants after two weeks, I forwarded the initial email with a brief follow-up message inquiring as to whether they had had a chance to review my request for participation. If after the follow-up reminder I did not hear back after one week, I followed up with a telephone call. In one instance, an individual on my recruitment list contacted me after having heard about my study through a colleague.

Once participants agreed to participate we negotiated a time and place to meet for the interview. Prior to our meeting I emailed the study consent form in case they wanted to review it in advance of our meeting (see Appendix I: Consent Forms). Given the provincial scope of NBS I conducted interviews across the province at a range of venues. I interviewed participants at locations of their choosing including offices or conference rooms at their place of business, their homes, coffee shops, and food courts in hospitals or malls. In three instances I interviewed participants over the telephone due to scheduling conflicts and prohibitive travel costs.

Data “saturation” is a concept frequently used among qualitative researchers to defend their sample size and decision to cease data collection (Merriam, 2009: 219). Essentially, the principle is that the further one gets into the data collection, the researcher will “begin to hear the same things over and over again, and no new information surfaces” (Merriam, 2009: 219). However, this concept of “saturation” is contested on the
grounds that such a claim is a “somewhat arrogant assumption that one has tapped all relevant human variation” (Thorne, 2008: 161). Recognizing the limitations of claiming saturation, I ceased recruitment once consistent themes were evident across participant categories. In addition, my decision to end recruitment was further confirmed once participants stopped recommending new names, suggesting that I had recruited a significant number of the key players engaged in the expansion and implementation of NBS in Ontario.

**Interview Data Collection**

**Interview Data**

Interview data for this embedded case study analysis of perceptions of consent for NBS in Ontario were generated from 57 interviews. The interviews, which were audio-recorded and transcribed verbatim, produced approximately 1400 pages of transcript data.

**Interview Guide**

Given the exploratory nature of my inquiry I conducted semi-structured interviews – considered appropriate for case study research (Hancock & Algozzine, 2006) – which were comprised of open-ended questions, taking the form of a guided conversation (Merriam, 1998). Since this study was conceptualized from the outset within a public health ethics framework, the interview guide was, accordingly, developed to align with public health ethics principles (see Appendix J: Interview Guide). Drawing on Kass’ (2001) public health ethics framework I designed questions to elicit participants’ perceptions regarding the goals of NBS, who benefits from NBS, and who bears the burdens of NBS. I also asked participants whether they thought the goals of NBS were changing in light of an expanding
program that now discloses carrier status for sickle cell anaemia, for example, and whether this expansion fits within the mandate of NBS. I also incorporated a question in my interview guide to elicit participant perceptions of consent for NBS in Ontario. As I have previously indicated in Chapters 2 & 3, the issue of consent for public health interventions such as NBS is a topic of great debate not only in NBS scholarship, but also in the public health ethics literature. Regarding consent for NBS in Canada, and Ontario more specifically, in 2001 Wildeman and Downie challenged Canada’s – and Ontario’s – implied consent approach to NBS on legal grounds. Also in 2001, Ontario’s Provincial Advisory Committee on New Predictive Genetic Technologies recommended express written consent for NBS and, in the event that the province opt for an alternative to express consent, a committee should be established to examine and defend alternative propositions. Given the debate in Ontario, as contextualized within the broader international debate on this topic, I was interested to learn what key informants connected to the expansion and implementation of NBS in Ontario thought about the consent debate.

In keeping with the exploratory nature of my study, I also wanted to create a space for new information, insight, and perspectives to emerge (Merriam, 1998). Therefore, the very first question on the interview guide was designed to put participants at ease and build rapport as well as provide participants with an opportunity to share information which they perceived to be important and free from my construction of the issues at play (Merriam, 1998). Questions similar in design asked participants to speak to what they perceived to be the most pressing issues facing NBS in Ontario and areas that they would like to see changed or modified moving forward.
Interviews

As previously noted, I met participants for the interview at their office, home, coffee shop or food court. In three instances, interviews took place over the telephone. Some participants had read and signed the consent form prior to my arrival and others had not (see Appendix I: Consent Forms). In all cases, prior to initiating the interview, we reviewed the informed consent form together and I asked participants whether they had any questions. In addition to requiring a participant’s signature, I had two questions embedded in the consent form on the signature page in the form of a check box. The first question asked whether participants consent to having their name associated with the facts related to the development of Ontario’s NBS program and the second whether they would consent to a follow-up interview in the event that clarification or elaboration was necessary. Many participants inadvertently skipped these two check-box questions and went straight to signing the form, returning only after I drew their attention to the two questions.

After we reviewed the consent form, I asked participants once again whether it would be permissible to audio-record the interview. Once I received their permission, I turned on my recording devices and proceeded with the interview. All except two of the 57 interviews were audio-recorded. One was not recorded due to a technological malfunction. The second interview was not recorded because at the time, the interaction was more an informal conversation – with me taking comprehensive notes – rather than a formal study interview. However, at the end of the conversation the participant wanted to sign a consent form to be considered a study participant. In both instances I took notes during the interviews followed
by post-interview memos. Interviews ranged in length from 20 minutes to 3 hours with the average interview lasting 60-75 minutes.

**Transcription**

Each interview was transcribed in full and as close to verbatim as possible. I transcribed half of the interviews myself and arranged for two transcription companies to complete the remainder. I edited all of the transcripts completed by the companies to address typographical and spelling errors, dialogue errors, missed dialogue, and to insert italics, underlines, and bold fonts to capture voice intonation and emphasis.

**Interview Data Description, Analysis, and Interpretation**

Merriam (1998; 2009), like Stake (2005), maintains that qualitative case study analysis and interpretation can draw on a wide range of analytic and interpretive approaches. With respect to qualitative analysis in case study research, Stake (1995) asserts that there is no “right way” to analyze data. He insists that, “each researcher needs, through experience and reflection, to find the forms of analysis that work for him or her” (Stake, 1995: 77). Similarly, Margarete Sandelowki (2000) espouses that “qualitative work is produced not from any “pure” use of a method, but from the use of methods that are variously textured, toned, and hued” (p. 337). Kvale and Brinkmann (2009) underscore that much of the influential qualitative research has taken many forms with respect to their analytic strategies, ranging from strict and structured to more informal and eclectic approaches.

Given the analytic and interpretive freedom afforded researchers engaged in qualitative research generally and case study inquiries in particular, I conducted a three-tiered
approach to my qualitative case study, transforming my data through description, analysis, and interpretation (Wolcott, 1994). However, the qualitative description, analysis, and interpretation dimensions of a qualitative inquiry occur simultaneously and cannot be parsed out in a linear fashion (Wolcott, 1994; Stake, 2005; Merriam, 1998):

By no means do I suggest that the three categories – description, analysis, and interpretation – are mutually exclusive. Nor are lines clearly drawn where description ends and analysis begins, or where analysis becomes interpretation. (Wolcott, 1994: 11)

Still, I have tried to separate the process as much as possible for clarity and transparency. In my findings chapters six and seven I distil the sections according to descriptive themes and analytic/interpretive themes. The final stage of this inquiry (see Chapter 8) involves an applied public health ethics analysis that draws on Public Health Ontario’s (2012) public health ethics framework, *A Framework for the Ethical Conduct of Public Health Initiatives*.

**Description**

The goal of first order inquiry is to understand the descriptive dimensions of the phenomenon (Sandelowski, 2000). As Wolcott (1994) states, “*Description* addresses the question, ‘What is going on here?’” (p. 12). Qualitative description is critical for the general understanding of the case and to serve as the backdrop for the analysis and interpretation (Wolcott, 1994; Sandelowski, 2000). I began with “generic coding” (Thorne, 2008: 147) with a combination of pre-established codes and codes derived inductively from the transcripts (Stake, 1995; Merriam, 1998). For instance, such categories as “consent”, “storage and research”, “carrier status disclosure”, “goals of newborn screening” were general coding structures that mirrored my interview guide, but were also solidified as
legitimate headings during the transcription process. Other coding categories, however, were generated as I read the transcripts. Such inductive codes included “education”, “HCP knowledge”, “comprehensive program”, and “incidental findings.” During this first phase of coding I made notes in my thesis journal about themes, new insights, and possible interpretations of the data.

After I conducted the first phase of general coding, I sorted all of the general headings within a transcript together in a Pages document. Each document contained a header containing the category code and the participant identification number. Upon completing this first phase of general coding, I identified consent as among the most pervasive topics discussed by participants within and across transcripts and categories. While I did have two explicit questions about consent in my interview guide, a number of participants introduced the topic independently. In addition, many of the ethical issues introduced and discussed by my participants during the interviews such as education, awareness, carrier status disclosure, and storage and research converged with the ethical issue of consent. Given that qualitative case studies need to be “flexible” and “responsive to changing conditions of the study in progress” (Merriam, 1998: 8), I decided to focus my analysis and interpretation on the policy and practice of consent for NBS in Ontario. Moreover, the clear tensions that the topic of consent evoked within and between participants in terms of how best to achieve the public

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26 The transcription process was critical to my analysis. While transcribing the interviews or editing the transcripts, I would recognize themes within or across transcripts. In such instances I would type a transcription note directly into the transcript using brackets (e.g. [TRANSCRIPTION NOTE: ………]). In addition, I would write transcription memos in my thesis journal, which was housed in a Bento database and categorized under the journal heading “Transcription Analysis Notes.” Through the act of memoing I began to identify emerging themes, contradictions, and new ways of thinking about my data. As a result of the transcription process the basic narrative thread became visible and the structure for my thesis findings began to take shape.

27 Pages is the Macintosh equivalent of Word.
health goals of NBS – namely identifying and treating affected infants – while also promoting parental autonomy seemed worthy of a deeper inquiry.

*Analysis*

The analysis stage of the inquiry provides insight into “how things work”: “Analysis addresses the identification of essential features and the systematic description of interrelationships among them—in short, how things work” (Wolcott, 1994: 12). Once all of the codes were aggregated, I engaged in a second iteration of coding which was more explicit and specific (Thorne, 2008: 147). This phase of coding served to generate themes within the overarching category of consent. I drew on grounded theory analytic techniques such as focused coding and constant comparative analysis (Merriam, 1998; Charmaz, 2006; Thorne, 2008). I also identified emerging themes through what Kvale and Brinkmann (2009) refer to as “meaning condensation.” Meaning condensation is the act of synthesizing long interview passages into a few thematic words or phrases (Kvale & Brinkmann, 2009). I applied this process to the aggregated data generated through the general coding phase. After reading the transcript data I identified “the natural ‘meaning units’ of the text, as they are expressed by the subjects” (Kvale & Brinkmann, 2009: 205). I then consolidated the main theme of each meaning unit as succinctly as possible to facilitate the deeper investigation of how the identified themes related to one another and the larger research question at hand (Kvale & Brinkmann, 2009).

To facilitate this process, I created a series of two-column tables. Each emergent theme had its own table. For instance, my analytic tables served to explicate such themes as “challenging informed consent”, “challenging implied consent”, “confusion and consent”,


“consent as fiction”, “implied consent as inherited”, “logistics of consent”, “risks/barriers/harms of consent”, “education and consent”, “HCPs and knowledge”, and “fear of refusals”. At the top of the table I wrote a brief explanation as to what the theme encompassed. In the right column of the tables I inserted the raw data and in the left column I wrote analytic memos (Merriam, 1998). These analytic memos served to situate the excerpt within the broader context of the interview narrative and to deconstruct the text, looking specifically at participant language, assumptions, and arguments. Through the process of memoing I developed new insight and understanding which led to the creation of new themes, tables and memos.

As the “transformation of qualitative data” (Wolcott, 1994: 10) progressed, codes were collapsed to reflect the more interpretive dimension of the inquiry (Thorne, 2008). For example, I ultimately characterized participant perceptions of consent that fell under the themes of “parental refusals”, “risks/barriers/harms of consent”, “logistics of consent” and “HCP knowledge” within a broader framing of “consent causing harm.” Such themes as “consent as fiction” and “challenging informed consent” became the theme of “consent as ritual.” And participants’ perspectives on “the check box” and calls for “a simple consent” and “decision aids” comprised the thematic heading “reconceptualizing express consent.” This final stage of meaning condensation results in an analysis upon which to base the interpretation (Kvale & Brinkmann, 2009).

**Interpretation**

Interpretation is the final stage of the data transformation:
Associated as it is with meaning, the term interpretation is well suited to mark a threshold in thinking and writing at which the researcher transcends factual data and cautious analyses and begins to probe into what is to be made of them. (Wolcott, 1994: 36)

Having identified the broader analytic themes pertaining to implied and express consent for NBS I then interpreted these themes within my public health ethics lens. As I outlined comprehensively in Chapter 2, I interpreted these themes of consent within the broader public health principles of least restrictive alternatives, autonomy, effectiveness, and social justice. In addition, the interpretation of my data in relation to the public health ethics principles worked to underscore the perennial challenge faced by public health and public health informed programs, namely to find a balance between the rights and freedoms of the individual and the collective health of the public. In my chapter on express consent, for example (see Chapter 7), I took the analytic themes of “consent causing harm”, “consent as ritual”, and “reconceptualizing express consent” and interpreted them within the polarizing public health tension.

For example, I interpreted the analytic theme of “consent causing harm” within the broader public health concerns of causing harm to infant health and by extension population health. On the other side of the tension, I used that analytic theme “consent as ritual” to illuminate perceived harms to autonomy if consent for NBS were introduced. (I recognize that individual and collective interests are not always mutually exclusive and that there is necessary overlap when considering benefits and harms. And while I acknowledge this overlap as it pertains to my analysis and interpretation, I think that for clarity purposes there is usefulness in presenting my analysis and interpretation in a way that mirrors the public health tension that contributes to the foundation of public health ethics.) Since a goal of
public health ethics is to find a balance between the two poles – protecting individual rights and freedoms on the one side and protecting broader societal health interests on the other – I interpreted the analytic theme “reconceptualizing express consent” as an effort to consider what consent should look like within a public health context such as NBS that would work towards narrowing the gap between individual and collective interests.

Applied Public Health Ethics Analysis

Throughout the processes of description, analysis, and interpretation, I noticed that many of the participants in my study waged a series of critiques against the current practice of implied consent for NBS in Ontario and the possibility of express consent for NBS in the future. For instance, in this study, participants discussed the perceived benefits and risks of consent, the potential strengths and weaknesses of consent, and ways in which the benefits could be enhanced and the potential harms minimized. While the ideals of an express consent policy for NBS were lauded (even among those participants most adamantly opposed to express consent for NBS), concerns about the implementation of consent in practice were raised by many participants who discussed consent (even among those participants who felt that a more formalized consent policy was necessary). Such perceptions of consent led me to interpret the practice of informed consent as a least restrictive alternative for a public health program that warranted critical analysis. Given the potential for least restrictive alternatives to cause harm (Guttman & Salmon, 2004; Nuffield Council on Bioethics, 2007; Faden & Shebaya, 2010) I decided as the final dimension of this inquiry to apply Public Health Ontario’s (2012) public health ethics framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, to the practice of consent for NBS.
As I explained in Chapter 2, Public Health Ontario’s (2012) public health ethics framework, *A Framework for the Ethical Conduct of Public Health Initiatives*, is a framework designed to assist professionals engaged in public health initiatives think through the public health ethics dimensions of a specific program, intervention, policy, or practice. As I have argued, given the public health roots of NBS in Ontario, this framework is useful as a lens through which to examine and analyze the policy and practice of consent for NBS. Drawing on my data and the existing literature on consent for NBS more broadly I offer an applied public health ethics analysis of the practice of consent. This exercise illuminates the perceived benefits and harms of consent for NBS in a comprehensive way, underscoring that the concerns of express consent for NBS extend well beyond fears of parental refusal. Appreciating the multi-dimensionality of concerns around consent is critical as the advisory committee evaluates Ontario’s current consent policy moving forward. By considering the perceived strengths and weaknesses of consent for NBS through a comprehensive public health ethics lens allows for a proactive approach towards minimizing these potential harms.

**Interview Data Presentation**

To capture and represent a dynamic interview in a text-based form, I relied on familiar transcription notation (Sandelowski, 1994; Poland, 1995; McLellan *et al.*, 2003). Specifically, I used ellipses to capture the evolution of my participants’ train of thought including stuttering, changing their mind with respect to word choice, starting sentences anew, and in some instances mini-pauses. In the illustrative quotations presented in the findings chapters that follow, ellipses in brackets ([…]) signify that the quotation is either starting in the middle of a larger piece of dialogue or is omitting a passage between the
featured text in order to maintain the focus of the quotation. Text that appears in square brackets signifies either a clarification with respect to prepositions or to substitute identifiable proper nouns with more general nouns to protect participant anonymity. Parentheses are used to contain interviewer text (e.g., interviewer’s questions, probes, responses, words of encouragement), to capture pauses and other descriptors pertaining to intonation (e.g. participant says excitedly, participant bangs on the table), as well as background noises. Italics are intended to capture words or phrases that were stressed or emphasized in the dialogue and bold font captures a level of expression above the intonation reflected through italics. In some instances, words will appear in all capital letters and/or underlined to underscore that the person speaking was extremely emphatic.

Although the interviews were transcribed as close to verbatim as possible (Poland, 1995), to render the featured quotations easier to read I removed unnecessary stuttering that did not contribute meaningfully to content as well as such interviewer interjections as “um hm”, “right”, “okay”, “yes” and “yeah.” These interjections interfered with the flow of the quotation and did not advance the meaning or purpose of the quotation. In the few instances where I was not able to hear the audio clearly, I have written in parentheses “inaudible” and if I think I heard the words, but not with 100% confidence, I have included “sounds like: what I think I heard” (e.g., (inaudible: sounds like “shouldn’t we try”)).

**Rigour and Trustworthiness**

The “non-formulaic complexity of the qualitative research process” (Eakin & Mykhalovskiy, 2003: 190) has led to much debate among qualitative scholars as to the appropriateness of checklist criteria designed to appraise qualitative research (Sandelowski,
1986; Murphy et al., 1998; Yates, 2003; Eakin & Mykhalovskiy, 2003). Some qualitative researchers have endeavoured to augment “procedural considerations with a ‘substantive’ orientation that centres on the relationship between research practices and substantive findings and interpretation” (Eakin & Mykhalovskiy, 2003: 192). Yates (2003) argues that with small-number research, issues of selection and comparison are important but an emphasis on techniques of data-treatment and comparison is misplaced because the meaningfulness and potential contribution of such studies lie in acts of interpretation and dialogue with the broader field. The more such studies move to mimic factor-analysis procedures, the more they undermine their claim to be any more than a report on ‘a small sample’. (p.224-225)

However, purposive sampling, trustworthiness of transcription, fairness, auditability, peer debriefing, reflexivity, and the transferability of findings are among the accepted ways that rigour and trustworthiness of a qualitative inquiry can be established (Sandelowski, 1986; Murphy et al., 1998; Yates, 2003; Eakin & Mykhalovskiy, 2003). These are the criteria that best capture my efforts to establish rigour in my study on perceptions of consent for NBS in Ontario. Since I followed Merriam’s (1998; 2009) approach to qualitative case study inquiry, whereby the researchers can draw on a host of available qualitative analytic and interpretive techniques, adhering to these established elements of rigour and trustworthiness is appropriate.

**Purposive Sampling**

Selecting the appropriate sampling strategy for the inquiry is part of establishing study rigour (Baxter & Jack, 2008). I implemented two purposive sampling strategies to increase the likelihood of identifying participants who were either key players at the time of the program’s expansion and/or critical to its current implementation: maximum variation sampling and snowball sampling. The goal of maximum variation sampling is to allow for
the possibility of diverse perspectives on a given issue (Thorne, 2008):

When selecting a small sample of great diversity, the data collection and analysis will yield two kinds of findings: (1) high quality, detailed descriptions of each case, which are useful for documenting uniqueness, and (2) important shared patterns that cut across cases and derive their significance from having emerged out of heterogeneity. (Patton, 1990: 172).

I purposively recruited participants from three groups: advisory committee members, HCPs at treatment centres, and advocates. Snowball sampling contributed to the recruitment of individuals in these three groups in addition to leading to the creation of a fourth category of participants, namely “legal experts”.

Trustworthiness of Transcription

Transcript data was central to this qualitative description, analysis, and interpretation. The quality and trustworthiness of the transcription contributes to the rigour of a qualitative inquiry (Poland, 1995). Transcripts are constructed texts and transcription is an interpretative activity (Sandelowki, 1994; Poland, 1995). Decisions that researchers make about what to include in a transcript “directly influence the nature and direction of analysis” (Sandelowski, 1994: 311). Furthermore, in the process of transcribing, “[j]udgment calls are often required in interpreting what should be committed to paper” (Poland, 1995: 298), particularly decisions pertaining to sentence structure and construction (Poland, 1995). I made the decision at the outset that I wanted my transcripts to reflect as closely as possible the live interview. Therefore, each transcript captures all verbal utterances made by both the interviewer and the interviewee including stammers, repetition, hums and hahs, “crutch words” (e.g., you know), laughs, coughs, sneezes, audible breaths, et cetera (Sandelowski, 1994; Poland, 1995). In addition, to the best of my ability, I captured on paper voice
intonation, inflection, emphasis, volume, “mimicking voices” (Poland, 1995: 298), and
accents through the use of italics and bold font, underlining, all capital letters, and
parenthetical notations such as (says in a high pitched voice) or (says in a whisper) or (says
with a smile), *et cetera*. I also noted pauses and interruptions between the interviewer and
the interviewee. I also documented in each transcript non-verbal sounds (e.g., participants
bangs on the table) and gestures (e.g., participants makes an “okay sign” with their fingers)
acquired through the audio recording as well as drawing on field notes from the interviews
(Poland, 1995).

I transcribed approximately half of the interviews myself. Although I hired
transcriptionists to aid in the transcription of the remaining interviews, I painstakingly
reviewed all of the transcripts against the recordings to address what Poland (1995) describes
as deliberate alterations of the data (e.g., misrepresentations of the audio recording),
accidental alterations of the data (e.g., sentence structure, mistaking words, omissions, the
use of quotation marks), and unavoidable alterations of the data (e.g., non-verbal cues, voice
inflection, body language). Since I conducted all of the interviews and was heavily involved
in the transcription, the transcript data that served as the basis for my analysis are as faithful a
representation of the interview as possible. Still, I recognise the inherent interpretive
dimension of transcription and the inevitable judgment calls exercised, particularly with
respect to sentence construction.

*Fairness*

Fairness, defined as the extent to which all “stakeholder views, perspectives, claims,
concerns, and voices” (Lincoln & Guba, 2000: 180) are represented in the text is also among
the criteria used to appraise qualitative research (Murphy et al., 1998; Lincoln & Guba, 2000). Failing to include the views of certain stakeholders is perceived as a kind of bias (Lincoln & Guba, 2000). I have incorporated the perspectives from all four groups represented in my study sample and worked to ensure that I provided original data from as many participants as possible.

Auditability

The “auditability” of a qualitative inquiry requires a clear portrayal of the decisions made throughout the study so that another researcher can follow the logic and could come to similar conclusions “given the researcher’s data, perspective, and situation” (Sandelowski, 1986: 33). Auditability is typically established in the research account itself (Sandelowski, 1986). Therefore, I have provided in this thesis a faithful, descriptive account of the evolution of this inquiry, the theoretical lens that I have used throughout the analysis, how the study was conducted, and the turning points in my analysis that led me to make the analytic and interpretative connections that I made. In addition, in the chapters that present my findings (Chapters 6 & 7) I have provided raw data to enable readers to engage with a small subset of my data. Ultimately, however, I have offered one interpretation of my data presented in a way that endeavours to enhance the readers’ capacity to ‘feel’ the texture of the account being put forward, to understand the conceptual development and foundations of the analysis and, thereby, ultimately, to better apprehend the leaps of imagination and creative thinking that constitute (arguably) the most valuable feature of all research, qualitative or otherwise. (Eakin & Mykhalovskiy, 2003: 191-192)
**Peer Debriefing**

Peer debriefing or peer examination, whereby colleagues are asked to reflect and comment on the research inquiry and the emergent findings, also contributes to the rigour of a qualitative inquiry (Merriam, 1998; Murphy *et al*., 1998). Peer debriefing can take many forms and I have incorporated a number of peer debriefing avenues into my research over the course of this inquiry.

First I have presented my findings at various stages in “workshopping” sessions designed to engage peers unfamiliar with my work in dialogue and to receive feedback on my study design, methodological approach, analysis, and interpretations and to gain additional insights as a result of receiving fresh perspectives and insights on my data. In addition, I have given numerous national and international presentations on my work to audiences both within and outside my disciplines of public health and bioethics. Presentations are a key component to the data discovery process (Lincoln & Guba, 2000) and through these initiatives I received much constructive feedback that contributed to the evolution of my inquiry. Finally, my thesis committee contributed to the peer debriefing process as well by providing feedback on my work at various stages and in various capacities, challenging me to think through ideas, connections, interpretations, and style of presentation. Collectively these exercises in peer debriefing at various pivotal junctures throughout this inquiry served as a form of checks and balances and ultimately contributed to the confirmation that the logic and interpretive connections ultimately made fit solidly within the theoretical lens and data with which I was working.
Reflexivity

Reflexivity is another critical dimension of establishing rigour and trustworthiness in a qualitative study (Merriam, 1998; Murphy et al., 1998). Qualitative inquiry requires that the researcher engage in critical self-reflection to “assess the likely impact of the researcher’s presence on the data obtained” (Murphy et al., 1998: 188). In considering my position as a researcher within the context of an exploratory qualitative case study of the perceived ethical issues connected to NBS from the perspectives of key informants deeply connected to and invested in NBS in Ontario, my background as a qualitative researcher in bioethics definitely contributed to the way in which I was perceived and received by some of my participants.

For a number of participants affiliated with the treatment centres, my position as an ethics scholar was arguably perceived as an asset in that participants were eager to share with me their perceptions of the ethical issues that they saw as needing to be addressed in order to enhance the experience of NBS for parents. For many of the advocates who participated in my study, particularly those engaged in advocacy work for individuals and their families living with sickle cell anaemia, I was welcomed warmly and perceived as another avenue through which their message could be disseminated. For others, particularly among past and present members of the advisory committees, my academic location in ethics in particular appeared to elicit initial reactions of scepticism in terms of my personal motivations for conducting this research. At various time points in the interviews, participants’ unflattering perspectives about bioethics, ethics research, and bioethicists emerged:

I think the flames are fanned a little bit by our ethicists (participant laughs). I think, you know, to be blunt about it [inaudible 45:15 s/l: sometimes], they need a job (participant is still laughing) and they need to study these things. But the risks attached to it (pause) are not always as… as large as we would think. (#15, advisory committee)
[Informed consent for NBS is] Only [a big debate] in a narrow circle of academics … and in a slightly broader circle of people who haven’t thought seriously about it, with respect. (#2, advocate)

There’s nobody been suing people for doing a heel prick on their baby and there’s no human cry that this is not right, except from the bioethicists (participant laughs, smiling). (#5, advisory committee)

I’m a sceptic when it comes to ethical research. (#18, advisory committee)

So I think it’s the paranoia that is, and mass hysteria around it [privacy], and it’s become an industry unto itself. (#16, advisory committee)

For a couple of participants, the qualitative nature of my inquiry generated scepticism as well. Qualitative research was perceived by one participant as “naval gazing.” Another participant disclosed to me that the nature of the interviews too closely resembled journalism, which consequently affected what this participant felt she or he could share with me.

Despite the scepticism exhibited by some participants, I incorporated what I interpreted and experienced as judgment into my analytic strategy. Specifically, I made consent within the context of NBS the “villain” in order to challenge the often taken-for-granted assumptions of consent as a presumed good. By incorporating this “villainous lens” into my inquiry and then contrasting it with participants’ more positive notions and understandings of consent, they worked in concert to identify, describe, analyze, and interpret participants’ tensions between parental autonomy and NBS uptake. This approach allowed for an interpretation of my data that hopefully serves as a foundation from which future efforts can be made to craft consent policy and practice within a public health context such as NBS that will work to narrow the gap between the interests of individuals and those of a population.
My positionality both as a student researcher and, for some participants, a fellow researcher engaged in NBS-related research also influenced my inquiry. I noticed a very interesting trend among a handful of participants: when they began to speak about the government they wanted me to turn off the audio-recorder. While some participants insisted upon it, others made the request that I turn off the recorder and then recognized what they had asked and then changed their mind. I cannot say whether my student status contributed to this instinct or if a more established researcher would have encountered the same phenomenon. However, I interpreted this reaction as a sign of a lack of trust—a fear that I would inappropriately handle their “data” despite having outlined the safeguards and measures I had undertaken to protect my participants’ information and identity.

As for my role as a fellow researcher engaged in NBS-related research, a focus of a number of my participants’ academic work as well, a couple of participants disclosed a perceived conflict of interest. As a result of this conflict, one participant openly confessed that this affected his or her contributions in the interview. Although only one participant openly stated that it affected the comprehensiveness with which he or she answered the interview questions, given the number of participants with overlapping interests, this may have been a factor in other interviews as well — though perhaps unconsciously done. I return to this point in the study limitations of this thesis.

Finally, the last reflexive insight that I think is important to acknowledge is my position as an “outsider” for all of the groups that I interviewed. While this may have contributed to the lack of trust among some individuals, the outsider position most distinctly affected the content of my interviews with members of the NBS advisory committees. These
participants had all signed confidentiality agreements in relation to their advisory committee work. Therefore, they were not able to speak freely about the issues and debates discussed in the committee meetings. Given that I was not part of this group, I was not privy to certain information, which necessarily affected the kind of data I was able to collect on the issue of consent for NBS. Specifically, I was able to attain only personal perspectives rather than personal perspectives in relation to larger group discussions.

Transferability of Findings

“Transferability” (Merriam, 2009: 224) and “extrapolations” (Merriam, 2009: 225) are two concepts that refer to the extent to which qualitative study findings can be transferred to other contexts or the extent to which one might extrapolate from one study and apply it to another situation (Merriam, 2009: 224). Both approaches rely on the reader of the study to make such connections more so than the researchers themselves, as the reader will be more able to assess similarities and differences between the case presented and the case for which the findings may be applicable (i.e., the policy and practice of consent for NBS in other jurisdictions) (Merriam, 2009). However, in order for readers to make such connections, the researcher must employ rich description to facilitate such an assessment. In addition, within the context of a single, exploratory, qualitative case study, the goals and objectives are to “understand the particular in depth, not to find out what is generally true of the many” (Merriam, 2009: 224).

With respect to this study, my findings are grounded in key informant perceptions of the policy and practice of consent for NBS in Ontario. However, the ethical issues related to consent for NBS are arguably not unique to NBS in Ontario. Therefore, while my analysis
and interpretation of participants’ perceptions of consent for NBS are specific to Ontario and contextualized further within Ontario and Canadian health law, the public health ethics lens integrated into the study design, analysis, and interpretation offers the possibility that my findings may be transferrable to other NBS programs facing similar decisions about consent: “As Wolcott (2005, p. 167) points out, ‘every case is, in certain aspects, like all other cases, like some other cases, and like no other case’” (cited in Merriam, 2009: 228).

**Ethical Considerations**

Three research ethics boards (REBs) in Ontario approved this study (see Appendix K: Research Ethics Board Approvals). The University of Toronto was the first REB to approve this study. Many of my participants were public figures appointed to various advisory committees while also affiliated with hospitals and/or universities across the province or individuals working for the NBS treatment centres. The University of Toronto’s Office of Research Ethics would not guarantee that their REB approval covered the ethical recruitment of these individuals and, therefore, despite the minimal risk nature of the study, I was advised to contact all of the hospitals and universities connected to my participants to see whether I needed to undergo any additional REB reviews. After speaking with individuals at each of the Offices of Research Ethics, two required that I go through their review: Queen’s University and the University of Western Ontario.

During the course of my study two ethical considerations emerged. One became visible through the snowball sampling recruitment process and the other pertained to protecting participant anonymity in the presentation of my data. Given the relatively small NBS community in Ontario, snowball sampling raised an ethical issue that I had not
anticipated. I found myself temporarily troubled in terms of how to invite potential participants identified through snowball sampling to join my study. Some participants were clear that I could use their name when initiating contact with the individuals they recommended I recruit for my study. However, I felt that disclosing my source would not only reveal their participation in my study, but could conceivably place undue pressure on my invitee. Ultimately I decided to use a variation on the phrase, “Through my research to date, I have learned that you played a role in the expansion of newborn screening in Ontario” in my recruitment email. However, some participants recruited through this strategy wanted to know who recommended them at the time of the interview. In those instances where the participants had given me their permission to use their name I disclosed my source. In one instance I said that I was not able to say due to confidentiality and as a result it temporarily upset the rapport and dynamic of the interview. Similar challenges emerged at the end of interviews when I asked, “Is there anyone else with whom you think I should speak?” Participants willing to offer recommendations often wanted to know who I had already spoken with so as to be most helpful by not making repeat suggestions. A common response to the aforementioned question was, “Have you spoken to so-and-so?”

Protecting the anonymity of participants in the presentation of the data is considered a challenge in case study research in general (Merriam, 2009) and in my case study in particular given the individuals involved in NBS in Ontario is relatively small and some participants expressed explicit concern that they might be identifiable. Although each quotation in the findings chapters is presented anonymously and in aggregate within the context of four constructed categories (advisory committee, treatment centre, advocate, and
legal expert), certain turns of phrase, idioms, manner of speaking, and/or grammatical errors could potentially render a participant identifiable. Therefore, to minimize this risk I removed grammatical errors, proper nouns, and any other language that could possibly reveal participant identity from the featured quotations. As a further precaution I asked one of my committee members, Dr. Fiona Miller, who is closely connected to individuals involved with NBS in Ontario, to try and put a name to the featured quotations. In the few instances where she could identify my participant through the quotation, I took measures to anonymize the quotation more effectively.

Chapter Conclusion

In this chapter I introduced my approach to case study research, defined my case, and explained the analytic, interpretive, and applied techniques I used to provide new insight into a longstanding debate on the issue of seeking parental consent for NBS. I also articulated the measures I took to ensure the rigour, trustworthiness, and ethical integrity of this inquiry. The chapters that follow demonstrate through the presentation of my findings how I transformed my data through description, analysis, and interpretation. Chapter 5 describes my four categories of participants and Chapters 6 and 7 document my participants’ perceptions regarding implied and express consent for NBS in Ontario. Chapter 8 is a chapter devoted to an applied public health ethics analysis of consent for NBS.
Chapter 5. Participants

Chapter Overview

This chapter introduces the four general categories that comprise the 57 voices featured in the subsequent findings chapters. To protect the anonymity of my participants, I present my findings in aggregate using four constructed categories: advisory committee, treatment centre, advocate, and legal expert. These categorical distinctions are useful in outlining the different roles my participants play (or played) in the larger NBS in Ontario narrative. It is important to underscore, however, that the boundaries of these categories are fluid, with many participants assuming multiple roles. For example, a participant serving as an advisor on a NBS committee may also work at one of the treatment centres and/or be engaged in various advocacy initiatives. For the purposes of this study, however, I have categorized participants according to only one role. With the exception of the legal experts in my study, if participants who served on an advisory committee (past and/or present) could ultimately be placed in more than one category, I classified them according to their role as an advisory committee member. This was a deliberate decision based on the Maternal-Child Screening Committee and Newborn-Child Screening Sub-Committee’s mandates to address issues pertaining to consent for NBS. I thought it would be useful to offer the perspectives and attitudes of past and/or current committee members as the most recent committees continue to debate these issues. Similarly, if individuals working at the treatment centres

28 If legal experts fit more than one category, I decided to keep their contribution distinct and explicitly defined as lawyers and/or legal scholars as I recognize that their legal background presents the possibility of providing a unique perspective grounded in their understanding and interpretation of the legal parameters of consent and should be differentiated from the other category perspectives.
were also engaged in NBS-advocacy initiatives, I grouped them within the treatment centre
category given that their professional experience addressing the outcomes of NBS would
offer a particular perspective, likely distinct from those of both the advocates and advisory
committee members.

**Participant Demographics**

**Advisory Committee Members**

I recruited 24 participants who serve or served on one or more of the advisory
committees (past and/or present) established to address emergent issues pertaining to NBS in
Ontario (see Table 3 below). Members of these advisory committees over the years have
included – and continue to include – individuals with professional interest, expertise, and
commitment to NBS. In addition they bring expert knowledge in endocrinology,
hematology, biochemistry, metabolic genetics, pathology, health policy, and ethics.

**Health Care Providers and Other Professionals Working at Newborn Screening Treatment
Centres and Newborn Screening Ontario**

I interviewed 14 individuals from across the five regional NBS treatment centres as
well as HCPs and other professionals working at Newborn Screening Ontario. Participants
included in this cohort included nurses, genetic counsellors, physicians, laboratory
technologists, and administrators (see Table 3 below).

**Advocates**

I also recruited 15 advocates to participate in my study. Of these 15 participants
whom I have classified under the label “advocate”, 12 were patient-, parent-, community
organization-, and other independent advocates; three were HCP-advocates (see Table 3). The majority of these participants advocate on behalf of individuals and their families living with hemoglobinopathy disorders, sickle cell anaemia and thalassemia in particular. Other advocates represented in my sample include those supporting individuals and their families living with inborn errors of metabolism and cystic fibrosis. Some participants were champions of the general expansion of NBS in Ontario. These advocates who participated in my study were largely recruited through snowball sampling. In an effort to differentiate the advocate voices from one another without compromising participant anonymity, quotations from HCPs will be identified as “HCP-advocate”—the others will be identified as “advocate”.

Legal Experts

I also recruited four legal experts as a result of my snowball sampling strategy. These participants comprise a mix of legal scholars and/or lawyers both within and outside the province. Participants who recommended that I interview these individuals in my study perceived these legal experts to have contributed in some way to the debate on consent for NBS in Ontario.
### Table 3: Participant Categories

<table>
<thead>
<tr>
<th>Participants</th>
<th>Number of Participants</th>
<th>Category Label</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Members of government advisory committees addressing newborn screening in Ontario</strong>&lt;sup&gt;1&lt;/sup&gt;</td>
<td>24</td>
<td>Advisory committee</td>
</tr>
<tr>
<td>▪ Advisory Committee to the Ontario Ministry of Health on Screening for Inherited Diseases in Children&lt;sup&gt;2&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Advisory Committee on Inborn Errors of Metabolism&lt;sup&gt;3&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Provincial Advisory Committee on New Predictive Genetic Technologies&lt;sup&gt;4&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Newborn Screening Subcommittee of the Ontario Advisory Committee on Genetics&lt;sup&gt;5&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Ontario Advisory Committee on Newborn and Childhood Screening&lt;sup&gt;6&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Ontario Children’s Health Network (OCHN) Newborn Screening Impact Task Force&lt;sup&gt;7&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Advisory Committee on Newborn and Childhood Screening’s Task Force on Inborn Metabolic Disease and Special Diets&lt;sup&gt;8&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Maternal-Child Screening Committee and Newborn-Child Screening Sub-Committee&lt;sup&gt;9&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Better Outcomes Registry Network&lt;sup&gt;10&lt;/sup&gt;</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Health care providers and other professionals from one of the 5 regional treatment centres and Newborn Screening Ontario</strong></td>
<td>14</td>
<td>Treatment centre</td>
</tr>
<tr>
<td>▪ London Health Sciences Centre–Children’s Hospital of Western Ontario</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Hamilton Health Sciences Centre/McMaster University Medical Centre</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ The Hospital for Sick Children</td>
<td></td>
<td></td>
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<tr>
<td>▪ Kingston General Hospital</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Children’s Hospital of Eastern Ontario</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Advocates</strong></td>
<td>15</td>
<td>Advocate</td>
</tr>
<tr>
<td>▪ Patients</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ Parents</td>
<td></td>
<td></td>
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<tr>
<td>▪ Community organization representatives</td>
<td></td>
<td></td>
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<tr>
<td>▪ Independent advocates</td>
<td></td>
<td></td>
</tr>
<tr>
<td>▪ HCPs</td>
<td></td>
<td></td>
</tr>
<tr>
<td><strong>Lawyers and legal scholars</strong></td>
<td>4</td>
<td>Legal expert</td>
</tr>
</tbody>
</table>

<sup>1</sup> Members of the Advisory Committee to the Ontario Ministry of Health on Screening for Inherited Diseases in Children, Advisory Committee on Inborn Errors of Metabolism, Provincial Advisory Committee on New Predictive Genetic Technologies, Newborn Screening Subcommittee of the Ontario Advisory Committee on Genetics, Ontario Advisory Committee on Newborn and Childhood Screening, Ontario Children’s Health Network (OCHN) Newborn Screening Impact Task Force, Advisory Committee on Newborn and Childhood Screening’s Task Force on Inborn Metabolic Disease and Special Diets, Maternal-Child Screening Committee and Newborn-Child Screening Subcommittee, Better Outcomes Registry Network.

<sup>2</sup> Further information on the Advisory Committee to the Ontario Ministry of Health on Screening for Inherited Diseases in Children can be found [here](#).

<sup>3</sup> Details about the Advisory Committee on Inborn Errors of Metabolism are available [here](#).

<sup>4</sup> Information on the Provincial Advisory Committee on New Predictive Genetic Technologies is [here](#).

<sup>5</sup> More details on the Newborn Screening Subcommittee of the Ontario Advisory Committee on Genetics can be found [here](#).

<sup>6</sup> Further information on the Ontario Advisory Committee on Newborn and Childhood Screening is [here](#).

<sup>7</sup> Details on the Ontario Children’s Health Network (OCHN) Newborn Screening Impact Task Force can be found [here](#).

<sup>8</sup> Information on the Advisory Committee on Newborn and Childhood Screening’s Task Force on Inborn Metabolic Disease and Special Diets is [here](#).

<sup>9</sup> More details on the Maternal-Child Screening Committee and Newborn-Child Screening Subcommittee are [here](#).

<sup>10</sup> Further information on the Better Outcomes Registry Network is [here](#).
Chapter Conclusion

My study sample is comprised of 57 participants. These participants offer their perspectives on and attitudes towards the policy and practice of consent for NBS in Ontario. Their perspectives are shaped in part by their experiences as advisory committee members, professionals employed at the treatment centres and Newborn Screening Ontario, advocates, and lawyers and/or legal scholars. In the subsequent chapters, I provide descriptive analyses of my participants’ perspectives on such topics as implied consent for NBS (see Chapter 6), and express consent for NBS (see Chapter 7) as well as offering an interpretation of these perspectives as examined through a public health ethics lens. The next chapter thematically explores participants’ perceptions of the current consent practice for NBS in Ontario: implied consent.

Notes:

1. The information presented on each of these committees was gleaned from a combination of participant interviews, follow-up correspondence with a select number of participants, as well as available press releases, reports, committee terms of reference, and related documents. Participants who discussed the various committees with me often expressed uncertainty around the specificity of some of the committee names and dates. Consequently, the information provided about each committee in the notes below may contain some inaccuracies. That said, the committee details, beyond establishing participants’ membership in these advisory capacities, were not central to the overall analysis of my inquiry given that confidentiality agreements prevented the substance of these meetings from being discussed in interviews. However, providing an accessible history of the evolution of the NBS committees since the program’s inception in the 1960s may be beneficial for documenting purposes and future research initiatives.

2. The Advisory Committee to the Ontario Ministry of Health on Screening for Inherited Diseases in Children was established in 1968 by Order in Council. This committee officially disbanded in 1991/1992. However, a number of the physicians committed to newborn screening, and involved in the five academic
centres in Ontario, continued to meet annually in an unofficial capacity under the new title, the Advisory Committee on Inborn Errors of Metabolism (participant communication, 2013).

3. The Advisory Committee on Inborn Errors of Metabolism succeeded the aforementioned Advisory Committee to the Ontario Ministry of Health on Screening for Inherited Diseases in Children in an unofficial capacity beginning around 1991/1992. This committee reported to the Ministry of Health and Long-Term Care through the Public Health Branch and operated until 2004 (participant communication, 2013).


5. The Newborn Screening Subcommittee of the Ontario Advisory Committee on Genetics was established in 2005 and, by the end of the year, had provided the Ministry of Health and Long-Term Care with its recommendation to expand the province’s newborn screening program (Ministry of Health and Long-Term Care, 2005a; 2005b).

6. The Ontario Advisory Committee on Newborn and Childhood Screening was formed in 2006 (Ministry of Health and Long-Term Care, 2006). This committee reported to the Ministry of Health and Long-Term Care until 2009 (participant communications, 2011), after which point it was replaced by the Maternal-Child Screening Committee and Newborn-Child Screening Subcommittee (see Note 9 below).

7. The Ontario Children’s Health Network (OCHN) Newborn Screening Impact Task Force was established in 2006.

8. The Advisory Committee on Newborn and Childhood Screening’s Task Force on Inborn Metabolic Disease and Special Diets was established on March 31, 2006 and remains active. Initially this committee reported to the Integrated Policy and Planning Branch of the Ministry and then transferred to the Public Drug Program Branch a few years later (participant communication, 2013).

Booth, 2012). On April 5, 2010 and April 30, 2010, a call for Expressions of Interest was circulated to initiate committee membership selection processes (Born Ontario, n.d.a). An announcement made in 2010 describes the creation of three committees: The Maternal-Child Screening Committee and the Prenatal and Newborn-Child Screening Sub-Committees (Born Ontario, n.d.a). The first committee meetings were held in 2010 and the committees remain active. The Prenatal and Newborn-Child Screening Sub-Committees report to the Maternal-Child Screening Committee and PCMCH. The Maternal Child Screening Committee reports to PCMCH and Better Outcomes Registry and Network (BORN). PCMCH and BORN then report to the Ministry of Health and Long-Term Care. (see Born Ontario, 2013a; 2013b; 2013c)

10. Better Outcomes Registry Network (BORN) was established in 2010 after the Ontario Perinatal Surveillance System was granted registry status (Peel Public Health, n.b.a). BORN reports to the Ministry of Health and Long-Term Care—Maternal Child Youth Health Strategy (Born Ontario, 2013a).
Chapter 6. Perceptions Of Implied Consent For Newborn Screening In Ontario

Chapter Overview

Implied consent is the current approach to consent for NBS in Ontario (Hayeems et al., 2009; Miller et al., 2010b; Bombard et al., 2012). In Ontario, implied consent is considered a form of informed consent whereby individuals must be informed about the type of treatment, the benefits and risks of treatment, the alternatives to treatment, and the potential implications of not having the treatment (Health Care Consent Act, 1996). Implied consent, as explained earlier in the introductory chapter of this thesis, “occurs when surrounding circumstances are such that a reasonable person believes that consent has been given, although no direct, express or explicit words of agreement had been uttered” (College of Physicians and Surgeons of Ontario, 2006: 5). However, debate persists as to whether implied consent is appropriate for patients generally (College of Physicians and Surgeons of Ontario, 2006) and for NBS specifically (Wildeman & Downie, 2001; Provincial Advisory Committee on New Predictive Genetic Technologies, 2001).

Within the context of my interviews the topic of consent was introduced through a variety of avenues including discussions about perceived parental awareness about NBS, parent and HCP education, carrier status disclosure through NBS, storage of and research on blood spot data and samples, and more directly about the approach to consent for NBS in Ontario specifically. During the interviews many participants offered their assessment of
both implied consent for NBS in Ontario and express consent, a possible alternative (see Chapter 7).

In this chapter I begin by presenting descriptive themes that work to establish that at the time of the NBS program’s expansion in 2006, the issue of consent was addressed in the advisory committee meetings. Given that these meetings were confidential, I was not able to learn specifics about the nature of the discussion, debate, official reasoning behind the Ministry’s decision to maintain the implied consent approach for expanded NBS, or what recommendation the advisory committee gave to the Ministry (see Chapters 4 and 9 for more on this). However, participants shared their personal perceptions as to why the implied consent model was pursued for expanded NBS in Ontario. I labelled these descriptive themes “Discussion Around Consent for Newborn Screening Pre-Program Expansion” and “Perceived Justifications for Implied Consent for Newborn Screening in Ontario” respectively.

The remainder of this chapter is devoted to exploring participants’ personal attitudes towards implied consent for NBS in Ontario. Independent from participants’ perceptions about why they believed the decision was made to continue with implied consent, many participants had their own views about implied consent for NBS in the province. I identified three dominant analytic/interpretive themes that raise questions about the appropriateness and effectiveness of implied consent for NBS in Ontario: 1) Challenging the concept of implied consent; 2) Perceived parental knowledge and understanding; and 3) Perception that parents need to be better informed about newborn screening.
Descriptive Themes

Descriptive Theme 1: Discussion Around Consent for Newborn Screening in Ontario Pre-Program Expansion

Consent was a topic of interest among many participants in this study. Although questions about consent were integrated into my interview guide, many participants brought up the subject independently. However, participants who served on the NBS advisory committees disclosed that they had signed confidentiality agreements. Therefore, they were limited in what they could discuss, particularly, it seemed, when it came to issues of consent:

You know I can't really get into that because there's legal advice um and there … there are legal issues there that … and it involves a lot of legal advice that we went to get from Counsel about what the analysis and the choices were made and that one I really can't get into. […] but um there's no question… the quest… it [consent] was looked at […]. (#57, advisory committee)

Despite these confidentiality restrictions, participants who served as advisory committee members were able to articulate their general perception that the issue of consent had been discussed at length prior to the expansion of Ontario’s NBS program:

**Interviewer:** I was wondering if you could talk about how the advisory committee came to decide that the implied consent model should be used?

**Respondent:** Ummm, that was a big discussion actually (participant says laughing), and, you know, do we go ahead with it and so the negative option kind of thing? Like if you don’t want it, then fine you don’t get it, otherwise if you don’t say anything we’ll go ahead and do it, and that’s essentially the way we progressed. (#25, advisory committee)

I think there was a lot of discussion. I think there was considerable discussion of how one would mandate newborn screening. (#42, advisory committee)

Um another issue that ah came up was related to um the use of the secondary use of the blood spots: the storage of them; the availability of them to researchers; um consent issues related to both those things. (#1, advisory committee)
I think the same sort of debates that we’ve had in this conversation [referring to the interview] have been in the advisory committee, so the same issues about whether should it [NBS] be mandatory with people having different opinions, um … how much consent is needed […] should everybody just be informed, should there be a choice about that, is there harm related to that, and … and um, I think there’s a bit of debate, which I think is healthy, and it’s not like there’s a right answer; so it’s not like I can say … I mean people … I think there’s healthy debate and I think that’s … I think it’s important for healthy debate to inform policy, so I think that’s a good thing about it. (#3, advisory committee)

It’s [informed consent was] certainly something that was considered very carefully um at the inception of the program. (#56, advisory committee)

While the consent policy was purportedly discussed, some participants on the advisory committee perceived the issue of consent for NBS as having been left unresolved:

Well we [the advisory committee] didn't come to a conclusion [on consent], that's one of the problems… (#54, advisory committee)

I don't know whether we went … we certainly discussed consent a lot. I don't think we made… we … the … I don't know whether we made a formal decision on it. (#15, advisory committee)

Others, however, felt an agreement on the issue of consent for the 2006 expansion of NBS in Ontario had been reached:

We [the advisory committee and Ministry of Health and Long-Term Care] were comfortable pursuing things the way we [Ontario] had always been doing them. (#57, advisory committee)

Although some of the participants in the advisory committees described discussing the issue of consent for NBS at length, they were clear that the ultimate decision as to what approach the province would adopt for NBS, whether mandatory screening, implied consent or express consent, lay not with the advisory committee or Newborn Screening Ontario, but rather with Ontario’s Ministry of Health and Long-Term Care:
Those are decisions that no advisory committee with a narrow mandate that we have can make. That’s a political decision that requires involvement of a... public representatives... elected representatives. (#54, advisory committee)

I don’t think it’s up to... it’s not up to me, it’s not up to us, it’s up to, you know, it’s up to the public as represented by the elected government to decide what is the appropriate consent framework for you know, um... um... for a public health intervention like this. (#18, advisory committee)

This is the other thing about the Advisory Committee is that they are really, truly, advisory only, and so they can make the recommendations to the Minister about what information should be given to families and what information should be on the website but they can take it or leave it depending on what their... their goals are in the moment. (#1, advisory committee)

We don’t... committees don’t make decisions on policy, I mean, the government makes those decisions but the people who are helping inform the government and helping raise the issues, I think it’s important to have a balance perspective. (#3, advisory committee)

One participant articulated that while the ultimate decisions lie with the Ministry, the Ministry looks to the advisory committee for guidance on such issues as consent for NBS:

[The] Ministry relies on this expert advisory group to give us their expert opinion on any given question related to newborn screening. (#56, advisory committee)

This theme underscores the belief among some participants who served as advisory committee members at the time of the program’s expansion that the topic of consent for NBS in Ontario was discussed prior to the implementation of the expanded NBS program. A few participants appeared to disagree as to whether a formal, conclusive recommendation had actually been put forth. Ultimately, however, a number of participants made it clear to me that while the Ministry of Health and Long-Term Care looks to the advisory committee for recommendations, the decision-making power lies firmly with the Ministry.
Descriptive Theme 2: Perceived Justifications for Implied Consent for Newborn Screening in Ontario

Although participants who served on the advisory committees underscored that policy decisions regarding consent for NBS were not theirs to make, they shared with me what they perceived to be the justifications for the continuation of implied consent for Ontario’s expanded NBS program.

The most common justification for implied consent for NBS in Ontario was an appeal to its tradition and history:

The essence was that…you know, the program is not new. It has been in place for 40 years (participant knocks on desk). So that’s the first thing: you have to acknowledge that we already have a program where … it’s … it’s in existence […]. (#15, advisory committee)

You know, we inherited a consent framework uh … we haven’t made any huge changes to it. (#18, advisory committee)

Like it’s always been that way, not just in Ontario, but really in Canada. (#20, treatment centre)

[I]t’s [NBS’s] been done for many years without informed consent so to speak […]. (#46, advocate)

Well, I think some of that was—I mean, this is jaded now, but that's always the way it was. From the time of Guthrie cards, it was just implied. So it probably just evolved into it's still just implied. (#31, advisory committee)

In addition to the “tradition” of implied consent for NBS in Ontario, a few participants felt that implied consent was even more justified now given their perception that parental education on NBS had improved significantly:
there was not a significant amount of discomfort in continuing to proceed the way we always had. Okay? Cuz there was not a change in terms of how it [the heel prick] was done before. And there is certainly um … ah *opportunities* should parents wish to um to not um have their child screened. Um and I think the … we were … I think physicians were… it was just something to be noted uh in the file, but nothing has… nothing changed. And in fact we felt that there was more information *now* out there to inform the implied consent piece. You know, like if they're quest… you know… so yeah informed implied. You know, there was … there was hardly anything out there before. (#57, advisory committee)

That other jurisdictions internationally had maintained a mandatory or implied consent approach for their expanded NBS programs provided some participants with additional reassurance and confidence that implied consent for NBS in Ontario was appropriate:

And, in fact, *most* jurisdictions in the western world do not require written consent. There are a few, but most of them don't so we were not *out of step* with the majority of in…[sniffs] in informing people and uh that they were… this test was going to be done, but then if they didn't want it they could opt-out, but they weren't going to ask *specifically* for uh consent or *assent*. (#54, advisory committee)

Other arguments put forth by participants appealed to the perceived collective agreement that expanded NBS is a “*worthwhile*” undertaking and “not a lot of push-back from people saying, ‘no, we should not be doing it’” (#25, advisory committee); “any parent in their *rational mind* would think this [NBS] was best for their baby” (#21, treatment centre); implied consent is “*practical*” (#24, treatment centre), “*much simpler*” (#37, HCP-advocate), “*formalized in law*” (#15, advisory committee); and, it “*seems to be working*” (#5, advisory committee). Some participants also underscored a lack of resources as further justification for pursuing an implied consent approach for NBS in Ontario: “I think you know, if we really had a lot of *resources*, it would be wonderful to sit down and explain everything. But, unfortunately we don’t have.” (#53, treatment centre)
Despite the perceived justifications of implied consent for the province’s NBS program, many participants offered their own perspectives on the perceived appropriateness and effectiveness of implied consent for NBS in Ontario. In the next section of this chapter I elucidate the three dominant analytic/interpretive themes identified in my data.

**Analytic/Interpretive Themes**

*Analytic/Interpretive Theme 1: Challenging the Concept of Implied Consent*

The concept of implied consent for NBS in Ontario has been challenged by scholars engaged in the debate on consent for NBS (Wileman & Downie, 2001). Some scholars insist that implied consent introduces the challenge of having to distinguish – in the absence of verbal cues – between parental consent and mere compliance (Wildeman & Downie, 2001). Even the College of Physicians and Surgeons of Ontario discourages HCPs from relying on implied consent as an approach to patient decision-making: “Although the Act states that consent to treatment may be express or implied, physicians are strongly advised to obtain express consent from the patient” (p. 5). Similarly, some of the participants in my study who discussed implied consent within the context of NBS in Ontario were equally wary of such an approach to consent. I identified three sub-themes that captured my participants’ struggles with and characterizations of implied consent for NBS in Ontario: 1) Should implied consent for NBS in Ontario be characterized as consent? 2) Is implied consent a voluntary or mandatory approach to NBS in Ontario?; and 3) For whom is implied consent effective?
Should implied consent for newborn screening in Ontario be characterized as consent?

Participants disagreed as to whether implied consent was the appropriate label to define Ontario’s approach to NBS. Moreover, some participants challenged the legitimacy of implied consent as a form of “consent”. One participant was adamant that implied consent is not consent, and, therefore, is the wrong term to describe Ontario’s approach:

**Respondent:** I think what they’re [Ontario’s] using is you can do it [NBS] unless they [parents] object, is that correct?

**Interviewer:** Yes. And that … it seems to be labelled implied consent, is that –

**Respondent:** That’s a big mistake.

**Interviewer:** Is it?

**Respondent:** Yeah, it’s not implied consent. There’s no such thing as implied consent. Um … it’s … it’s … when you start talking about implied consent, ah it’s really a fiction of consent and what it does is it confuses the situation where you have consent and consent is a justification for what you’re doing or you don’t have consent and you’ve got to find another justification for what you’re doing. And um (pause) so, I think that’s really important, that distinction. Basically, the courts have rejected implied consent. (#44, legal expert)

Participants in my study with legal expertise argued that introducing the language of consent necessarily requires that parents receive the information that would meet the legal and ethical standards of consent:

Well, you could call it something like an assumed permission model, that you … what you’re doing is you’re making an assumption that the parents have given permission, but they can, but it’s not consent, that’s the whole point. Once you use the … once you use the word consent, then you’re bringing in all of the requirements that are necessary, legally and ethically, for consent. So I can give you my permission to do something and in a loose lang… in a non-legal, non-ethical language, you would say, oh she consented to that. But if you looked at it legally or ethically, I haven’t cons… I haven’t consented unless I knew all about it, I knew the risks, I knew the harms, I weighed it up, I was the right person to do it, give the consent, you know, all that sort of stuff. So I just think you’ve just got to be careful about that. I think what you can do is you can describe a true consent, true, informed consent model, which is very clear and there’s no confusion...
about that. And you can describe a *true, compulsory or mandatory model*, which simply says there’s a law that lets us do this and, you know, we’re going to do it. And then *in between*, you’ve got *gradations* between those two poles. (#44, legal expert)

Implied consent is not the same as *no consent* […] You still have an obligation to make sure that people fully comprehend. (#14, legal expert)

Another legal expert explicates that implied consent *is* a legitimate form of consent within the healthcare context, but the extent to which it can be applied to such issues as storage of blood spots acquired through NBS, for example, remains unclear:

That was kind of the one *benefit* that the Privacy Legislation had kind of *clarified in the*… in… in… within… when you are doing something for healthcare *purposes*, you… you can *imply* consent. Like they *come to* the doctor um… with resp… and so you can *imply it*… as long as you’ve given them the information that they would need to… to… to… to object if they wanted to object. But the area you have to be *careful about* is that that’s… that's *where* you're using the information for healthcare purposes… like to *treat them* (participant raps on the table), to advise them about their condition, that kind of thing but where you get into some of these *other uses* for the information, the implied consent rules aren't so… it's… it's not so clear that you can rely on implied consent. (#7, legal expert)

Participants without legal expertise similarly questioned whether implied consent was the appropriate label to give Ontario’s current model. They maintained that in order to imply consent parents need to be sufficiently informed about NBS from the outset, a standard that they believe, based on their professional experience caring for infants who screen positive and their parents, is not being met (see also Analytic/Interpretive Theme 3: Parents Need to be Better Informed About Newborn Screening):

You could argue that implied choice is really not choice, I mean … that … that sort of implied choice … I mean what are the true elements of choice? So … if … if all we’ve done is given a pamphlet that very briefly outlines newborn screening, I mean I don’t think we’ve *really*, I don’t think that we’ve really *satisfied* the requirements of informed choice with that. (#3, advisory committee)
I’m not sure if that’s [implied consent’s] the actual term, the participant says in response to a question about implied consent, but that’s – I mean – just my perception […] I’m not sure if that’s the right term, I’ll have to think about it, I might change my mind (participant laughs). But um, basically I just know that families are not … like nobody’s sitting down and saying, “do you want to do this?” It’s more like, if you happen to know about it, you technically could say you don’t want it (participant says with a laugh). And you know, just being a genetic counsellor (participant says with a residual laugh) that’s not typically the way that we go through consents with families. (#52, treatment centre)

In addition to challenging whether the language of consent is appropriate for implied “consent”, a number of participants debated whether implied consent constitutes more of a voluntary or mandatory approach to NBS.

Is implied consent a voluntary or mandatory approach to newborn screening in Ontario?

Participants’ differed in their views as to whether implied consent is a voluntary or mandatory approach to NBS in Ontario. Some participants perceived implied consent as technically and legally a voluntary program given that there is no law mandating screening and parents are allowed to opt-out on behalf of their infants:

It’s [NBS in Ontario is] not mandatory; it’s voluntary. There’s no … In the States a lot of the States it’s mandatory by law. In Canada I think there’s one province where it’s mandatory. Maybe Saskatchewan but I’m not sure but all the other provinces it is voluntary. (#5, advisory committee)

Are they allowed to refuse? (participant laughs). You know, and yeah, of course they’re allowed to refuse. They have to sign the form, but … I’m not exactly sure why [parents would want to refuse screening]. (#33, treatment centre)

Yeah, I would have said it’s . . . it’s … yeah, well, it depends on where you stand, I guess, as to whether it’s mandatory or not. I think that option is always here to say ‘no’. Do we have families who do not want it [NBS]? Yes. You know. I mean, they don’t want to have Vitamin K, they don’t want to have screening and they only get ointment in their eyes because it’s mandated under the Public Hospital’s Act. So the law of the land says
‘thou shall have it done,’ but there is no law of the land that says ‘thou shall have your blood taken for a newborn screen.’ (#25, advisory committee)

Still, some participants questioned the difference between implied consent for NBS and mandatory screening. They challenged whether a program such as NBS could really be considered voluntary if parents do not know that the heel prick takes place:

I’m suspicious about implied consent. I don’t see implied consent as being that different from being mandatory quite frankly (participant laughs). Cuz again, because I don’t think that people often know that [infants are screened after birth] ... so if you don’t know that you have a choice then you don’t have a choice; something’s going to happen to you and you don’t know that you can make it not happen, so it’s pretty close to being mandatory. (#1, advisory committee)

I’m convinced it’s [implied consent’s] the same model [as the mandatory screening in the United States]. Because as a personal experience ... and I’m in the field so I know that newborn screening will happen so I’m listening and I’m waiting to see when the nurse will present the newborn screening option to me. And if they do either they’ve done it and now they’re telling me that they’ve done it, or they will say newborn screening will be done, but it’s so limited. And so, really I’m talking about personal, but I don’t know every hospital in the province, but I’m sure that they don’t spend enough time to explain exactly what they will investigate, and I’m sure that that has an impact. Because if a mom is getting a phone call from a physician saying that newborn screening is positive for something, I’m sure that they have no … they, most of them have no knowledge that they did something. (#17, advisory committee)

That a few participants inadvertently referred to NBS in Ontario as a mandatory program further illuminates the argument that the distinction between mandatory NBS and implied consent is – at least in practice – extremely nuanced if it exists at all:

**Respondent:** It’s [NBS’s] set up as a mandatory thing unless you object extremely. It’s applied to every birth in the province. It has the official support and integration of health care services and education programs and everything is being provided on a public basis. All CHEO is doing is acting as a contractor. *A really good contractor*, but a contractor.

**Interviewer:** Okay. (Pause) So when you give the label of a mandatory program, do you differentiate at all between mandatory and then implied consent, which seems to be the label that Ontario’s been...?
**Respondent:** Yes, it’s implied consent but it’s not *absolutely mandatory*. I mean, if you birthed your kid with your midwife or with your physician, wherever you were, and you walked out of the system and said, “You’re not doing this to my kid,” they couldn’t actually *force* it. I don’t think it’s a *legal statute* that you are *required* to provide the specimen. I forget the exact legal basis for creating the screening … the updated screening program, but I *don’t believe* that it mandates them, for example, to *force* blood collection at the point of police action, which I would see as mandatory (Pause). I don’t think it actually forces them, so I think it’s an implied consent […] I think it should be in effect something that’s *very high compliance* unless there is some *extreme* objection to which I cannot actually *conceive* of for this program, it’s not like you’re trying to *transfuse* somebody. It’s not like you are taking a life-threatening specimen. The specimen is taken as part of checking for jaundice and other accepted medical practice. It presents very minimal risk to the infant and it saves lives, so I don’t think it’s an issue. And I think the corollary is that *if you fail to make it mandatory* or *very high compliance* in some implied way *and you miss some kids* and they were diagnosed later, of *course you’d get sued for not* making it mandatory. So you can’t have your cake and eat it too. (#11, advisory committee)

Other participants discussed the voluntariness of implied consent for NBS by articulating their perceptions of the necessary requirements needed to meet the standard of implied consent — among them, knowledge and understanding of the intervention in question. Some participants – especially the legal experts – were adamant that parent understanding is directly relevant to conversations around implied consent for NBS. The information that parents receive and the extent to which they understand that information was perceived by some participants as fundamental to implied consent and essential to being able to assess “*whether it's reasonable* to presume that somebody consents” (#14, legal expert):

But *still* it [implied consent] means though … consent must be *informed*, consent must be given *voluntarily* ah … information … (thought trails off) So one of the things that will also be determined is the information that is given sufficient for the type of information that a reasonable person in the same circumstances would have wanted to know. (#14, legal expert)

Within the context of NBS in Ontario, one advisory committee member simultaneously argued that parents do consent to the heel prick in Ontario (underscoring the
voluntariness of the program), otherwise the intervention would constitute battery, but that parents do not fully comprehend the disorders for which their infants are being screened:

They [parents] obviously do consent to the heel prick, but I mean do they really … otherwise it would be assault … but do they really know what the disorders are that they’re being tested for, and I don’t believe that they do (participant says softly). And I think that’s a huge challenge […] I only mean that it’s consent in that the nurse will ask the mother if it’s okay to go ahead with the heel prick, um and it would only be done with the mother saying “yes,” so it’s consent in that sense; babies aren’t whisked out of the mother’s room with the heel prick, or anything like that. Um, and it’s not mandatory in the sense that if the mother said “no” they would not go ahead and do the heel prick. So that’s what I meant about consent in that sense. (#3, advisory committee)

This quotation addresses the perennial challenge of the practice of informed consent, whether express or implied: to ensure that knowledge and understanding underlie healthcare decisions. Moreover, according to the definition of implied consent (see *Health Care Consent Act, 1996*; Provincial Advisory Committee on New Predictive Genetic Technologies, 2001; College of Physicians and Surgeons of Ontario, 2006), failure to comprehend the details pertaining to the medical intervention performed ultimately compromises the consent given.

Despite the established criteria needed for a program such as NBS to feel confident that parental silence on the matter of NBS can be interpreted as informed consent, one participant articulated the belief that parent knowledge and understanding of NBS was distinct from implied consent and constituted “a whole other set of issues”:

So, the model in Ontario that we inherited was one where you … we … we imply consent from the health care provider who uh … uh … is taking the sample from the baby, right? Now, is the … you know, how many patients … how many kids … babies, um … you know parents have a full understanding of what was done, or you know, remembered that you know, what explanation did they get? Did they have a chance to express that whatever, right? That is … that’s a whole other set of … set of issues, and … and certainly education and knowledge in the public is something that you would want to see. (#18, advisory committee)
However, within the context of an implied consent approach to NBS in Ontario, public – and parent in particular – knowledge and understanding of NBS is necessarily connected to implied consent. For, as a few participants, particularly the legal experts, have articulated, NBS that occurs in the absence of consent – or some other established justification – is considered battery. One legal expert (participant #44) explains that in jurisdictions that have mandatory screening “you use the law and you authorize the screening through the law and that’s your justification” for the battery. In the absence of mandatory screening legislation, alternative justifications are required:

Now, there are alternative justifications. One justification is the required consent and in the case of a baby, that’s the consent of the parents. Another justification is what’s called legal authority, and that’s where the government passes laws or subordinate legislation, which are regulations that give the people the authority to do this, even if the parents don’t want it. And then a modified version of that is that you’ve got the right to … well you haven’t got the right … that you can do it, but there’s an exception if the parents object. You see, if you have a right, then people can’t object to you exercising your right, so legally we could … if … if … you can have an objection, you’d say that the healthcare professionals have got a privilege, a legal privilege, of doing this test, but that privilege is revoked, if … that’s how you would analyze it legally, if the parents object. (#44, legal expert)

Participant #14 (legal expert) underscores that battery occurs when a medical intervention occurs without having disclosed “the appropriate level of information”:

[I]f you conduct a medical procedure without complete ... without any appropriate level of information it actually constitutes battery which is ... which is a very significant ... tort; it is basically a kind of invasion of someone's physical integrity ... without any ... any ... without any consent is battery which is ... which is kind of serious ... serious tort. (#14, legal expert)

That some participants in my study debated implied consent as an approach to consent and disagreed as to where exactly it falls along the voluntary to mandatory spectrum suggests a level of skepticism and perhaps even a lack of confidence in the approach as a
form of consent — a lack of confidence shared not only by Wildeman and Downie (2001) but also the College of Physicians and Surgeons of Ontario (2006).

For whom is implied consent effective?

Of the participants who discussed implied consent for NBS in Ontario, I identified in the data a clear tension between participants’ perceptions of the goals of implied consent within a screening context and the goals of implied consent as an approach to consent.

Therefore, I divided this sub-theme into two parts, since participants talked about the perceived effectiveness of implied consent in two ways: 1) effectiveness as a universal newborn screening mechanism and 2) effectiveness as an approach to consent.

Implied consent as an effective universal newborn screening mechanism

Participants typically discussed implied consent in relation to its perceived role in getting infants screened. Phrases such as “it works” were commonly used to describe the implied consent approach for NBS in Ontario:

It [implied consent] works. They have 104% compliance. (Pause). They found 4,000 babies that they didn’t know existed every year. There are babies that are being screened who are not being registered. So the first year it started up they actually had 4,000 more kids sampled than they had birth records for. (#11, advisory committee)

More specifically, implied consent was perceived as a mechanism in service of a larger goal, namely the operation of an “effective screening tool” (#25, advisory committee) designed to identify sick infants expeditiously.

Implied consent as an effective approach to consent

However, participants also recognized that while they perceived implied consent as generating “greater pickup” second only to a mandatory program (#31, advisory committee),
their experiences with parents upon disclosing a positive screen suggests that implied consent within the context of NBS is less effective as an approach to consent:

Well, I mean, I think it's [implied consent’s] working well in getting babies screened. It depends on what—you know, working well for who or for what. I think it works well for getting babies screened because I don't know that the education is that great to … like to pregnant mothers, to learn really, fully about newborn screening, all the things that are testing for, you know, how that's followed up and that sort of thing. […] So, I mean, from that perspective, I don't know that you're really getting informed consent from the parents. But I think that model, the implied consent model, works well for getting all the babies screened because—(pause) because I don't think as many of the parents are truly informed about the testing, there's not a lot of people that are opting out of it, either. (#13, treatment centre)

Interviewer: Well it's considered … the model that has been adopted right now is "implied consent."

Respondent: Yeah. But the parents really don't. I can… from experience I would say that a lot of the parents know their kid's getting a blood test, but they don't know what test it's for. They aren't… its not explained, oh well you could pick up, you could be a carrier for CF and you might find that out. I don't think that's explained to them. (#34, advisory committee)

The extent to which the current approach to implied consent for NBS in Ontario meets “the standard of consent” (#3, advisory committee) was called into question, which I have interpreted as a challenge to the effectiveness of the practice of implied consent:

Well it bothers me that … um … all of these babies are having newborn screening testing and I don’t think parents really know what it is, so that bothers me, um, as health care providers […] So even within the present system, how could we inform parents better about what’s happening. So for me that stands out as something that we do better in other areas then we’re doing … of medicine … then we’re doing right here right now [with NBS]. Um … so I think the informing of health care providers and the informing of parents, um needs some improvement (participant says softly). Um … so … and I think the future concerns are much more around the storage and research [whispering], um, but right now I think we’re running a program with implied consent where it’s not … I don’t think we’re really meeting the standard of consent (participant says softly). So that kind of bothers me, I mean I think it’s … I don’t think we’re causing huge harm with that, because it’s … the conditions are rare, I mean I don’t think a lot of people are bothered
by it. I think it’s not truly what we would see as the gold standard, you know what I mean? So I think we need to try to do a little better. (#3, advisory committee)

This distinction participants made between the perceived effectiveness of implied consent for NBS as a universal screening mechanism contrasted against its perceived ineffectiveness as an approach to consent in terms of eliciting informed, autonomous, parental decisions mirrors the tensions articulated in public health ethics scholarship and the consistent challenge of finding a balance between accomplishing public health goals (e.g., achieving high screening uptake) while honouring individual autonomy (e.g., in the case of NBS respecting parental autonomy). Together these themes work to question whether that balance has been achieved through the current implementation of implied consent for NBS in Ontario.

Analytic/Interpretive Theme 2: Perceived Parental Knowledge and Understanding

The effectiveness of implied consent for NBS in Ontario as an approach to consent was challenged further by a number of HCPs in my study who question the level of parental awareness, knowledge, and understanding of NBS. Many of these HCP participants offered their perceptions, based on their professional experiences engaging with parents of infants who screen positive for a condition on the screening panel, of what parents in their care appear to know and understand about NBS in Ontario. While my participants cannot, of course, speak to parents’ actual knowledge and/or understanding of NBS in Ontario (and the findings below should not be interpreted as such), their perceptions of parental knowledge and understanding arguably inform their own views on the appropriateness and effectiveness of implied consent for NBS in Ontario.
Within this theme which I have labelled, “Perceived Parental Knowledge and Understanding,” I identified four sub-themes that highlight some participants’ struggles with implied consent: 1) Perceived parental awareness; 2) Perceived parental understanding; 3) Implied consent as a potential challenge to issues of social justice; and 4) Perceptions of parents’ knowledge regarding Newborn Screening Ontario’s policies on the storage of and future uses for dried newborn blood spots.

**Perceived parental awareness**

Participants involved in the direct care of infants and their parents who receive positive screen results perceived varied levels of NBS knowledge and understanding among parents in their care. According to the perceptions of participants who work in the treatment centres, some parents (upon learning that their infant screened positive for one of the conditions on the screening panel) appear to know that NBS had taken place, whereas other parents were characterized as having not known about NBS:

Maybe, I don’t know, *half the time* or so, maybe a little better than that [parents recall that their infant was screened], but there are definitely *a good chunk* that don’t recall the test *being done* […] (#24, treatment centre)

From an actual experiential thing, um… the *patients* who have screened positive and that we’ve interacted with, a lot of them don't recall […] anything about the… the… hearing *any* information about the newborn screening program until they’ve screened positive. And some of them are, you know, were a *little*, you know, upset by that, but by and large at the end of, you know, the… the meeting with the genetic *counselors* and going through the whole thing, nobody was that upset by it, to be honest. We haven't had one case here where people were… there's one case where it turned out to be a *true* positive. And they… they had no recollection of being told that this testing was done. So they were a little upset with that *process*, but in the end, they were really glad that they got screened. (#12, advisory committee)
So once they’ve [the baby’s physician has] contacted the family [to alert them to the positive screen result], then I’ll have either the healthcare professional either call me and let me know they’ve talked to the family or have the family call me. Then I’ll speak with the family and find out how the baby is doing. Explain init … like do they remember having blood spots drawn. (Participant laughs) Sometimes they don’t … they didn’t even really know what it [NBS] was about. (#33, treatment centre)

Cuz what I’ve found is that families that come in here often don’t even know their baby had newborn screening done (participant says with a laugh). So that’s a huge um a huge thing. So they might have gotten some paperwork but it might have been given at a not so great time to read over it (says with a little laugh). Um, I know a lot of mums have said they’ve kind of been given it when they’re in the delivery room which is probably not the best time (participant says laughing). So I … I think that that … there’s a lot of different reasons why people are not getting the information, and they’re surprised by the call – ‘what … what test was that?’ and ‘when did that happen?’ (#52, treatment centre)

So in those cases [when the family physician does not return my message about the positive screen promptly] I'll just call the family directly to report the [positive screen] result to them, which can be trickier because then they don't know who I am. Some people are okay with that um… but I've had… certainly had parents that are not okay with that and don't know who I am, they don't know why I'm calling, and it just adds to their anxiety. (#32, treatment centre)

Most people don’t know the newborn screen is being done period. (#21, treatment centre)

That HCPs involved in infant and parent care post-positive screen perceive a number of parents to be unaware that their infants had undergone NBS suggests that, at the very least, further exploration is needed to evaluate whether it is reasonable to maintain the implied consent model as currently implemented for NBS in Ontario. For if, as some of my HCP participants suggested, some parents were not aware that their infants were screened, these parents arguably had not consented and their silence should not have been presumed to constitute informed consent.
Perceived parental understanding

Understanding is a critical component of informed consent whether implied or expressed. Some HCP-participants related that of the parents who recall that NBS occurred, they do not appear to know why NBS was conducted or the conditions for which their child was screened:

So I'll begin by asking them [the parents] how much they know about newborn screening and if anyone had discussed it prior to their baby's birth or at delivery. And most patients will vaguely remember some sort of discussion, but not many will really understand what it was actually done for. (#32, treatment centre)

And then I call the family, cold-call, call them directly, and generally the response from the family is good. So when I call them, I tell them who I am, where I'm calling from, remind them about the newborn screening, usually refer to it as the heel-prick test. And most of the time, a very high percentage of the time, the parent knows what I'm talking about. They recall the heel prick test. Although they may not know exactly what it was looking for, they at least recall the test. (#13, treatment centre)

Yes, they’re [parents are] familiar with the process, but they often think it’s PKU. “Oh, my baby has PKU.” They associate the new…in my experience (participant says slapping hand on chest) they associate the newborn screen panel as PKU. They do not have an awareness of the number of conditions that are screened for on newborn screening. And I continually have to clarify this as not PKU. (#51, treatment centre)

Awareness of NBS and understanding NBS are two distinct issues, yet both are critical to securing parental consent. If parents do in fact lack understanding of NBS, as suggested by some of my participants who interact regularly with parents whose infants have screened positive, their capacity to provide informed consent is compromised.

Implied consent as a potential challenge to issues of social justice

Some participants voiced their concerns around implied consent for NBS by appealing to their perception of inequalities connected to the approach. Specifically, a few
participants raised questions of justice and fairness given that not everyone is aware of NBS. Moreover, they assert that because existing information about NBS is predominantly written, certain parents are privileged. I have situated and interpreted this data within the broader context of social determinants of health and their associated implications for issues of social justice.

First, many participants were adamant that parents need to be better informed about NBS. One participant underscored the connection between lack of awareness and the associated challenge to implied consent by articulating that parents need to understand that their silence on the matter of NBS is interpreted as consent. Otherwise, the process is not “fair” to parents:

[P]robably a big (pause) problem with that [implied consent] right now is that people many times won't even know that they are giving consent. And so I think that for it… for it to be fair, I think we do need a lot more education before the babies are born so that people can even… can give this im… implied consent. (#32, treatment centre)

One might extend the usefulness of the language of “fair” and fairness to address what participants have identified as a distinct variation in knowledge and understanding among the parents in their care: Is it fair that some parents know their infants are being screened and others do not? How NBS information is currently conveyed in Ontario, for example, was considered by some participants to contribute to issues of fairness. Some participants articulated a perceived advantage to educated, literate parents given that NBS information is disseminated predominantly through written media. The participant quotation below explicates perceived literacy challenges within the current implied consent context and questions whether all parents can read and understand Ontario’s NBS brochure (see Appendix F):
[...] what about people who don’t receive it [the NBS brochure], what about people who aren’t literate, what about the, you know, difficulty of understanding or the opportunity to ask questions [participant says softly]. So … so I would argue we … we … you know it’s an implied choice but is it truly? You know, I mean I think you could say if you aren’t given the information for a true choice, is it right to do that? (#3, advisory committee)

Similarly, the current education initiatives transmitted through pamphlets, websites, and some committed HCPs were not considered universal in their reach:

[T]hey tell you there’s a lot of programs out there to help people, right. And this and that’s out there to help you. If you don’t know how to access it, you don’t know it’s out there. If you don’t read it in the newspaper that the government has … has this website that you can go to and you will get this and this information, they don’t tell you; hence, you don’t know how to look up for it, right. So, what I don’t know won’t hurt me, basically. […] [T]he only people that know that information are people, I guess, the ones who would read the Globe & Mail and there’s an article in the paper and they read up on it. It’s not the everyday person that would know all that stuff, right. (#28, advocate)

Such concerns about awareness, literacy, and information dissemination strategies support an interpretation of approaches to consent for NBS as having implications for social justice. If, for example, the way in which information is presented and disseminated for an implied approach to consent for NBS privileges certain members of society over others, then some parents are likely exercising their autonomy through informed decision-making and others are not. As explained in Chapter 2, self-determination is critical to individual well-being. Creating the infrastructure necessary to support and promote self-determination with regard to healthcare decisions is arguably among the primary objectives of implementing a least restrictive alternative such as implied consent for a medical public health intervention. Perceptions among some participants that implied consent policy and practice for NBS in Ontario may fall short of achieving that objective, particularly as it pertains to certain members of society, raises questions for further exploration not only about the effectiveness
of implied consent, but also the implications for social justice. While there are obvious limits on what my participants’ perspectives can say about actual parent knowledge and understanding, the important point that I would like to underscore for the purposes of my public health ethics inquiry of consent is the perceived social justice component of consent and the importance of attending to this dimension of consent processes in the development of consent policy and practice for NBS.

**Perceptions of parents’ knowledge regarding Newborn Screening Ontario’s policies on the storage of and future uses for dried newborn blood spots**

Implied consent for NBS in Ontario currently encompasses not only the newborn screen, but also the storage of dried blood spots and the use of those blood spots for de-identified research as well (Bombard *et al.*, 2012; Newborn Screening Ontario, n.d.f). Some participants expressed doubt that parents are aware of the storage and research components of NBS in Ontario, some participants working with the NBS treatment centres disclosed that they themselves were unfamiliar with Ontario’s NBS storage policies:

**Interviewer**: Well, because I know right now they’re [blood spots are] stored for 19 years.

**Respondent**: Okay. I didn’t know that.

**Interviewer**: Oh, really!

**Respondent**: Yeah.
**Interviewer:** Oh, okay.

**Respondent:** So, 19 years, ‘til they’re adults then.

**Interviewer:** Mm-hmm.

**Respondent:** And what’s the reasoning behind that? (#33, treatment centre)

**Respondent:** No, I haven’t really heard about it [the storage of blood spots in Ontario]. My opinion would be (participant slaps hand on the table) again, as long as individual rights are protected and *confidentiality* (participant raps the table) um … I would support it if it was again (participant raps the table) for the good of our society (participant says with a little laugh) and our *health care dollars*, if it was *fiscally responsible*. Um, I would support it.

**Interviewer:** And so, I think some of the challenges around um, this issue of storage … *Currently* it seems that it’s stored for 19 years here in Ontario.

**Respondent:** Oh is it?

**Interviewer:** Yeah.

**Respondent:** Wow! (participant says softly) (#51, treatment centre)

**Interviewer:** And um… in terms of um… I was hoping you would comment on your views with respect to the storage of newborn blood spots?

**Respondent:** *That* I don’t know… so I’m assuming because they are stored…

**Interviewer:** Well cuz they’re stored for 19 years.

**Respondent:** For 19 years? […] Well I haven't really… I haven't really thought of this. I wasn't really familiar with um… with what Ontario does. […]

**Interviewer:** So … so you weren't aware that the blood spots were stored?

**Respondent:** *Well* I knew they were stored. I didn't… but I really didn't know for how long. I didn’t really think … Yeah, like I knew they were stored for awhile. If you had asked me how long they were stored for I wouldn't be able to answer. Yeah. (#32, treatment centre)

**Respondent:** *Why?* [are the blood spots stored for 19 years?]

**Interviewer:** Why are they stored?

**Respondent:** Why are we keeping them for 19 years?

**Interviewer:** Right.

**Respondent:** Do you know?

**Interviewer:** You're actually asking me?

**Respondent:** Yes. (#31, advisory committee)
Similarly, some advocates revealed during the interview that they were equally unaware of the storage dimension of NBS:

**Respondent:** I don’t think they’ve made that [the storage of blood spots] public … to the public, like tell us any of that, right. So, I think if you let them [parents] know about it you might have some negative impact on that. It’s, “why are you saving their blood?” Why? Why? So um … that’s all stuff I don’t think we need to know (says and laughs). It’s like, if you decide to make it public, you’re opening Pandora’s Box, you know.

**Interviewer:** So … you weren’t aware that the blood would be stored?

**Respondent:** No, and I don’t care. Me personally, I don’t care. But I’m telling you right now, if you make it public you’ll have a hell-of-a lash on your hands. (#28, advocate)

I think I have a little knowledge that it is stored … for how long? I don’t remember how long. (#35, HCP-advocate)

While these findings cannot speak directly to parental knowledge and understanding on the issue of blood spot storage, the fact that some participants intimately connected to NBS in Ontario — whether through professional ties to the treatment centres or through advocacy work — are not familiar with blood spot storage policies or the rationale behind those policies, one can reasonably question the level of awareness of parents further removed from Ontario’s NBS program.

Analytic/Interpretive Theme 3: Perception that Parents Need to be Better Informed About Newborn Screening

As mentioned in the first part of this chapter, a few participants justified the continued use of implied consent for expanded NBS on the grounds that the NBS education component was perceived to have improved greatly with the program’s expansion:
It’s important that the parents know [about NBS] and they certainly um they certainly know more about it now, I mean we’ve basically papered the province with, you know, French and English language pamphlets, they’re being distributed at all kinds of points of access. There's stuff online. Um there's, you know, these fact sheets and, you know, in twelve languages. (#57, advisory committee)

Still, despite these information dissemination efforts, a major theme across many participant interviews was the perceived need for improved parent education; a theme arguably informed by participants’ perceptions that some parents are underinformed or uninformed about NBS in Ontario.

Direct questions about parent education were not embedded in my interview guide (see Appendix J: Interview Guide). However, I incorporated follow-up questions about education in some interviews to accommodate the direction of the interview as well as to build on themes identified in previous interviews. The issue of the perceived need for more parent education was often introduced in response to questions about what participants perceived to be the most pressing issues facing Newborn Screening Ontario today; areas of NBS where participants would like to see change; and in relation to questions about consent for NBS:

**Interviewer:** What would you say are some of the imperfections of the program?

**Respondent:** In Ontario?

**Interviewer:** Yeah

**Respondent:** Um … (pause) a lack of public accountability, a lack of public disclosure, and I think that harms the best interests of the program [inaudible 2:15:52: sounds like “sometimes”] because people forget, or never learn, or never hear about the thing [newborn screening]. (#2, advocate)
I mean I’m not criticizing the program, I think they’ve put up a nice website, I think they’ve done their best to try and educate with limited budget, but um … and I think health care providers are doing their best to try and put those pamphlets in what they’re handing out, and doing that kind of thing, but I think we need to … certainly especially if it’s going to expand anymore, and start producing more common results; you know, if people are going to start getting more and more results, we need to educate both sides much better about what it is um … so um … cuz I think that’s what people expect in medicine (participant says softly). (#3, advisory committee)

I think most people... most families that are pregnant only get to know about newborn screening when they receive the information saying, when your baby is born we’re going to prick the heel. I think we need to do more to educate the public that there's newborn screening for a whole host of metabolic disorders that can be lifesaving. (#6, advisory committee)

That's another thing actually for your previous question of things that newborn screening can look for to doing is um… education of parents and even healthcare providers. Um… because many many parents do not know much (participant laughs) about the newborn screening program so I think that is a big area to work on. (#32, treatment centre)

Within this broader thematic category of improved parent education I identified four sub-themes: 1) What parents should know about NBS; 2) Why parents should know about NBS; 3) NBS information should be commonplace; and 4) Recommendations for parent education outreach. Parent education is arguably a critical component of any least restrictive alternative for a medical public health initiative and integral to successful consent policies and practices in particular. The themes presented below describe what my participants want the public, and parents more specifically, to know about NBS in Ontario.

**What parents should know about newborn screening**

When participants discussed the disclosure of NBS information to parents within the context of express parental consent (see Chapter 7 for more on express consent for NBS),
there was disagreement regarding how much information to give to people (e.g., a comprehensive explanation of all 28 conditions versus a simple description):

Well, it’s [implied consent is] certainly very practical. I mean you don’t… it’s nice (participant laughs) not to have to talk to everybody individually and it makes sense in that you can’t get consent for every condition on the panel, right? Like if the panel [inaudible 53:45] there are 30 conditions, you can’t possibly explain them all, you can’t possibly explain the outcome of all of them individually but I think that it is a very doable, you know, task to describe the general principle of newborn screening which is, you know, to identify an infant and prevent and treat and that kind of thing. (#24, treatment centre)

However, when participants discussed parent education outside of the context of explicit discussions around express consent for NBS, many participants discussed their belief that parents should be well informed about all dimensions of NBS from the screen itself, to the storage of dried blood spots, and the possibility of de-identified research.

Participants felt that parents should know what NBS is, why it is conducted, the conditions for which infants are screened, the possibility of false positives, ambiguous results, and that some conditions might not have as effective treatments as others:

So I think for parents, um a brief description of why there is newborn screening; what are the conditions; um, what’ll happen; the process. Just kind of taking them through, you know, what’s going to happen so that that fear is taken away from them. And then, you know, to be honest, that yes, some disorders we may not be able to pick up on the newborn screen, so if your child is unwell or something it needs to go to the hospital. Similarly if um … um for some kids … for some disorders there may not be that kind of optimal treatment or something. So those kinds of things need to go into the education aspects. (#53, treatment centre)

I’ll tell you what I think would be important is that they [parents] know a little bit about it [NBS], that they’ve learned it in prenatal class, they’ve had the person talk to them one on one, give them the information and tell them we are going to test for 20 diseases in your child. There’s a recognized incidence of false positive so, if you get a call from the doctor or the hospital that your child has to be retested, the chances are still pretty good that there’s nothing wrong. I think that they can accept it more that way than if it suddenly comes out of the blue after they're home with their child and they have no
background knowledge of the *chances* of this being a true positive rather than a false positive. (#5, advisory committee)

In addition, many participants felt that parental knowledge and understanding of the storage and research dimensions was critical:

And I do think that we have to have sort of fairly *clear*, again, messaging around what happens to data, what happens to the samples, you know, who has access, who has not. But it also has to be done in a considered manner. (#15, advisory committee)

**Interviewer:** … And so I was wondering if maybe you could speak to *that* in terms of whether you envisioned those things being part of the information—

**Respondent:** (participant jumps in right away) education, *absolutely*. *Absolutely*. *Yes*. *Yes*. Yeah, there should be transparency [regarding the storage of and potential future uses for dried blood spots]. People have the *right* to understand what the whole process is *about* and, *really*, this is only my own personal opinion. (#46, advocate)

*Well, if they … if they’re going to store it for a certain amount of time, parents should know, you know, *this* will be stored for a certain amount of years (participant bangs on the table) and um … we might use it for research and um give them the benefits, let them read the benefits of *why* you store it, or what can be the benefit and give … because *information is power*, knowledge is power, and *once* you give them information, information is going to be used in a *logical way*. *So*, it’s very important. (#35, HCP-advocate)*

**Why parents should know about newborn screening**

Participants felt that parents needed to be educated about NBS to reduce fear and anxiety (“so that the *fear* is taken away from them” (#53, treatment centre)), particularly in case the parents are among those who will receive a call informing them that their infant screened positive for one of the conditions on the screening panel:

“My *personal* point of view would be I don’t think that they educate people enough about *what it is*. I think that *asking* or *telling* a mother just after she *delivered* (participant laughs) or a parent, I don’t think people are thinking *straight*, you know. […] I don’t think people have an awareness of all the different things that are being screened for. And you know, nine times out of ten, it doesn’t matter cuz you’re not going to get a
Parents fully educated about NBS were also perceived as having the necessary knowledge to be proactive and vigilant in the care of their infants, particularly as it pertains to NBS:

[T]here needs to be more education of parents regarding newborn screening because ah … not only for the sake of parents understanding that this is in fact a genetic test but also for issues of follow-up, because something that has come to light is, there have been many situations where a test was missed or the paediatrician never got the results and then, consequently, a few years down the road a child had been diagnosed with phenylketonuria or something else. Unfortunately, because early interventions weren’t started, there had been dire consequences. So, if parents were better informed as to this is what this test is about, then they might be more diligent in, you know, when they get to the paediatrician’s office saying: ‘Oh, by the way, did my child’s test results come in?’ You know, if the paediatrician didn’t bring it up, they would know to engage and say: ‘Hey’, you know. (#46, advocate)

Newborn screening information should be commonplace

A number of participants were adamant that NBS education should be continuous, delivered and received at various time points throughout pregnancy. These participants felt that parents should be so familiar with NBS by the time they give birth to improve the likelihood that parents both know about NBS and understand its goals and objectives:

So, you know, it [information about NBS] should … it ought to be information that is there early enough and often enough that it feels kind of commonplace I think to the couple so that they understand it and adjust to it and recognize its benefits. (#42, advisory committee)

Because in the final analysis, the more education you do up front, the less problems you have downstream. And so for example we've made the point that educating about newborn screening should be incorporated into all… any prenatal classes for women who are pregnant. I don't know the extent to which that's being done, but the whole notion is that by the time that a woman gets to the case room (pause) she's already heard about screening, she… the whole business of consent, understanding, acceptance, and so it becomes much easier. (#54, advisory committee)
Yes, yes so that's... so then... that is the only time you can say that people are informed because they understand. So even if they are giving consent you know that it's a well informed consent. There's always the question of how informed are people when they get consent, sometimes they don't understand what they're doing and by embarking on a good education program on the public, I think most people would say, “Oh, I know about that already” because they've been educated about that in the public. (#6, advisory committee)

**Recommendations for parent education outreach**

Given the emphasis many participants placed on NBS education for parents, they offered advice as to how to build on and contribute to education initiatives. Providing information that people can relate to was considered critical: “You can provide all the education material you want, if people don’t (pause) relate to it, then it’s not going to do what you think it’s going to do” (#54, advisory committee). In considering parent education outreach a multi-media approach was put forth as ideal:

I once had the occasion to advise a [company] on … on … an educational program to… to enhance awareness of a particular disease. This is a bit of a side bit: it illustrates the point perfectly. So they wanted…they said, “Well, we’ll go to this meeting, we’ll hand out brochures.” I said, “You’re wasting your time. It’s a total waste of time. You may get two percent of the people that you target who will be affected by that approach.” “So what do you suggest?” (the company asks). Well I say, "We suggest a multi… multimedia approach. You have to have written articles. You have to have brochures. You have to have speakers at meetings. You have to have uh… videotapes that people can carry away.” You know, if you rely on one thing, especially if it is written…it may… it’s not going to have an impact. We know that. I mean it’s not a dream, we know that. And yet people still think, “Well we told … we told somebody.” I could give you a million examples of where that has gone wrong. (#54, advisory committee)

I think, you know, a lot of things have been done, but maybe they need to be done on an ongoing way. Things like writing in those parent magazines, um newspapers, television, those things. And … and you know, every antenatal visit at least for the ones who are getting it, they should hear about it. (#53, treatment centre)

Community advocacy groups were also identified as an important – yet significantly underused – resource for educating parents about NBS. A few participants perceived
Newborn Screening Ontario as having limited resources for education outreach.\(^{29}\)

Consequently, they suggested reaching out to and partnering with community groups as a relatively inexpensive but purportedly effective approach:

[M]ost families that are pregnant only get to know about newborn screening when they receive the information saying, when your baby is born we’re going to prick the heel. I think we need to do more to educate the public that there's newborn screening for a whole host of metabolic disorders that can be lifesaving. So I think that needs to be looked at as well, but the education of the public is necessary, but even more with regards to sickle cell disease education of the at risk community. So, there are many opportunities that one can use: their community leaders, their advocacy groups, their churches and mosques and all these places where people from those communities they have associations from Caribbeans from Africa from all... So there are opportunities where information could actually be concentrated to make it more effective in [inaudible 47:04/sounds like 'person']. So people would be aware that these genes are common among these communities and that screening is available to adults by going to their family physician for a simple test and that newborn screening is here to stay and the implications of being a carrier. So [participant claps hands together] I think this can be done even in partnership with these advocacy groups and these community groups so that it's community-based and it’s far more effective because it's (slaps hands on lap) being done by people in the community and that's the recommendation of making to the Ministry: I think it would be far more effective (claps hands together). (#6, advisory committee)

You could do some things that would be fairly cheap. And to me, if you took the newborn screening centre in Ottawa and you aligned them with [...] patient populations and what [they’re] doing in outreach—align them with several of these associations and with some genetic counsellors. They could drive out the education for you fairly cheaply. I mean, it really could be done. (#41, advocate)

Then there are other things as well. There are different hospitals will offer information sessions or prenatal classes. There are all sorts of different ways that the information can get out. It’s just deciding, I guess, which is the most cost effective and which is, you know, how they are going to reach more people? (Pause) Then, of course, there are the different organizations out there that represent the different rare disorders that the government could be supporting these organizations’ efforts to educate both healthcare professionals and the public regarding the benefits of newborn screening for this particular disorder. If they actually have an information session or a pamphlet that

\(^{29}\)“And the centre in newborn screening there, does education, they try, but they have no mandate and they have no real resources for it. It's all they can do to run the program.” (#41, advocate)
speaks specifically to the issue of newborn screening for sickle cell, let’s say for instance, if they could actually do some work on their own within their own community. (#46, advocate)

Education is arguably at the foundation of the least restrictive approaches implemented to achieve public health goals. A number of participants in my study articulated their perception that the NBS knowledge base of HCPs and parents needs to be improved in order for informed consent to be an effective least restrictive alternative for NBS. Within the context of implied consent, if a significant number of parents lack the knowledge and understanding of the health issue for which a least restrictive alternative has been implemented (e.g., newborn screening), the very goal of a least restrictive alternative – to promote self determination and protect individual liberties – is arguably compromised. Regarding NBS, education is fundamental to parental knowledge and understanding and, by extension, implied consent. Participant perceptions of a perceived lack of parental knowledge and understanding suggests, at the very least, that focused attention should be paid to assess whether it is reasonable to presume informed parental consent from an absence of a NBS refusal.

Chapter Discussion

The mandate for the Maternal-Child Screening Committee and Newborn-Child Screening Sub-Committee (the current advisory committees for NBS in Ontario), includes addressing NBS issues pertaining to consent, storage, and future uses for dried newborn blood spots (Born Ontario, 2013a). Two Ontario-based studies (see Araia et al., 2012 and Bombard et al., 2012) have already contributed, directly and indirectly, to the debate on

30 Bombard et al.’s (2012) public engagement study also included participants from Montreal, Quebec.
implied consent for NBS in Ontario. A survey study conducted by Araia et al. (2012) provided information about the province’s implied consent approach indirectly by exploring maternal knowledge of and satisfaction with Ontario’s NBS program. These researchers found that while 89% of participants (n=750) “reported having heard of NBS” only 69% recalled receiving information about NBS (p. 3). Less than 15% were familiar with the storage of and future uses for the dried blood spots (p.3). Of the 712 mothers who responded to the survey questions designed to establish their understanding of NBS, “37% of the participants were considered to have ‘high knowledge’” (p.4). A knowledge question included in this survey was whether mothers were aware that parents have a choice as to whether they have their infants screened: “only 35% responded correctly” to this question (p. 4). Araia et al.’s (2012) study findings provide an empirical example of the arguable limitations of implied consent as currently practiced for NBS in Ontario. That a significant number of the mothers who participated in this study lacked knowledge about important dimensions of the newborn screen (for example, that they had a choice about whether to have their infants screened or what a false positive result means), coupled with the largely pervasive lack of awareness pertaining to the storage and research dimensions of the program, arguably raises the question of whether it was reasonable for HCPs at the time to imply consent for NBS from those mothers who participated in this study.

While Araia et al.’s (2012) study arguably introduces challenges to the effectiveness of the practice of implied consent for NBS as currently implemented in Ontario, the public engagement study in Ontario reported by Bombard et al. (2012) found that some study participants challenged the appropriateness of implied consent for the storage of and research
on dried newborn blood spots: “Some [participants] were concerned with the “blanket consent” for NBS that was used to imply consent for storage and research with stored samples” (p.243). While participants in Bombard et al.’s (2012) study disagreed on “the value and degree of consent for storage for research” they were unified in their view that parents need to be informed so that they may opt out should they so choose:

A theme that was persistent across respondents in discussions about storage was the need to inform parents that storage for secondary use occurred following NBS. Information was considered a necessary precondition to create some minimal opportunity for parents to opt out. (p. 243)

The findings from my study, which surfaced perceived challenges to the effectiveness and appropriateness of implied consent for NBS in Ontario contribute to these implicit and explicit challenges to implied consent as a least restrictive alternative for NBS in Ontario. Many participants in my study – across all four participant categories – challenged the concept of implied consent. They questioned both its legitimacy as an approach to consent and its appropriateness as the province’s approach for NBS. Much of my data that addressed implied consent from the perspectives of HCPs in my study was rooted in their perceptions of parental knowledge and understanding of NBS as gleaned from their professional interactions with parents whose infants screened positive for one of the conditions on the panel. Many participants who discussed implied consent felt that parents were either completely uninformed or under-informed (e.g., perhaps aware that their infant had been screened, but likely less informed as to why screening occurred). I interpreted participants’ perception of parents’ perceived lack of knowledge and understanding as contributing to their call for improved parent education initiatives on NBS.
Education is the foundation upon which a least restrictive alternative such as implied consent depends. More specifically, using such an implied approach to informed consent demands an ability for HCPs to be able to reasonably presume that parents know about NBS and understand the various components that comprise NBS in Ontario to feel as confident as possible that an absence of refusal can in fact be interpreted as informed consent (Wildeman & Downie, 2001). (It is this very presumption, however, that fuels critiques of implied consent, as some scholars would insist such a presumption often cannot be made (Wildeman & Downie, 2001).)

The perceived predominant approaches to NBS information dissemination, namely NBS brochures (see Appendix F), a comprehensive website, and a “tear-off” parent information letter on the NBS requisition form (see Appendix G), were perceived by some participants to privilege literate and otherwise socially advantaged parents: an approach to knowledge dissemination and consent perceived to present the possibility of fostering autonomous decision-making in some parents but not others. I interpreted these insights and concerns as reflecting the potential social justice implications of implied consent. For if parents lack knowledge and understanding, they have not “given” consent. If parents have not “given” their consent, then they have not exercised their autonomy. If parents have not exercised their autonomy, then I maintain that, as a least restrictive alternative, implied consent arguably fails in its effort to reach a balance between promoting individual rights and freedoms and achieving its public health goals of having all infants screened.

The act of applying a public health ethics lens to the least restrictive approach of implied consent for NBS, underscores perceived strengths and weaknesses of such an
approach and creates an opportunity to assess and remedy possible shortcomings so that the Ministry of Health and Long-Term Care and Newborn Screening Ontario can feel confident that implied consent is attaining a true balance between respecting the autonomy of all members of society while also achieving the population health goals through high NBS uptake. Drawing on my public health ethics lens of inquiry, I would argue that an ineffective approach to implied consent for NBS (defined as an approach to consent that does not lead to informed, autonomous parental decisions) has implications for parental autonomy, social justice, and ultimately challenges the effectiveness of implied consent as a least restrictive alternative for NBS.

The public health ethics analysis and interpretation I provide of implied consent based on my data, and as situated within the existing body of scholarship on implied consent for NBS in Ontario, arguably raises two questions worthy of further empirical investigation: 1) Is implied consent for NBS a least restrictive approach for some Ontarians but subtly coercive in its efforts at non-coercion for others?; and 2) Is it reasonable to interpret parental silence as informed consent for NBS in Ontario today?

**Conclusion**

Many participants in my study articulated their perception, often based on their professional interactions with parents, that parents in Ontario are largely unaware or under-informed about NBS. Even a subset of HCPs working at the NBS treatment centres and a few advocates familiar with Ontario’s NBS program revealed their own personal lack of knowledge regarding Newborn Screening Ontario’s storage and research policies.
Collectively these findings work to support my participants’ call for improved public, parent, and HCP education on NBS and likely inform their perceptions of implied consent for NBS. Participants identified what they perceived to be the strengths and weaknesses of implied consent for NBS in Ontario: an approach that succeeds in achieving high screening uptake, yet is perceived to do so at the expense of parental autonomy. These attitudes towards implied consent reflect the enduring public health ethics challenge of finding a balance between protecting and promoting population health and respecting individual rights and freedoms. Supported by the findings of two Ontario-based studies on NBS whose findings elicit similar concerns about the appropriateness and effectiveness of implied consent for NBS in Ontario as currently practiced (Araia et al., 2012; Bombard et al., 2012), I maintain that one can reasonably posit that there is reason to believe that implied consent is currently not achieving this balance. Despite participants’ criticisms regarding the theory and practice of implied consent for NBS in Ontario, express consent as an alternative was by no means considered a problem-free solution (see Chapter 7: Perceptions of Express Consent for Newborn Screening in Ontario).

31 Participants’ criticisms focused largely on the framing of Ontario’s current approach to NBS as “consent”.

32 Although Araia et al.’s (2012) study did not address the implications of their findings for implied consent in the province, their findings that underscore maternal knowledge on NBS can arguably be used to support my arguments regarding the effectiveness of implied consent in the province.

33 Independent from whether implied consent achieves the public health ethics balance, further empirical exploration may also be necessary to assess whether implied consent for NBS as currently implemented in Ontario meets the required standard of implied consent.
Chapter 7. Perceptions Of Express Consent For Newborn Screening In Ontario

Chapter Overview

Most of Canada’s NBS programs operate using an implied consent approach to NBS (Wildeman & Downie, 2001) and the vast majority of states in the United States have mandatory NBS programs (Mandl et al., 2002; Bombard et al., 2012). However, a number of jurisdictions internationally have begun to shift towards express consent approaches for NBS (e.g., France, Germany, United Kingdom) (see Appendix A). Proponents of express consent argue that mandatory and implied consent approaches are increasingly less defensible given the expanded nature of the programs; the significant increase in the number of conditions for which infants are screened; the disclosure of carrier status; the storage of newborn blood spot data and samples; and the use of the stored blood spots for research (Ross, 2010). Arguments supporting mandatory screening programs emphasize the public health nature of the screening program (Faden et al., 1982; Nijsingh, 2007).

At the time I designed my interviews, the issue of consent for NBS was topical in the academic literature. In addition, the advisory committee established at the time of the expansion (Ontario Advisory Committee on Newborn and Childhood Screening) was “established to provide oversight and ongoing advice to the government on its newborn screening program” (Ministry of Health and Long-Term Care, 2005a) regarding its

34 In the province of Ontario, express consent is defined as an approach to informed consent whereby patients and surrogate decision makers must directly and explicitly give oral or written consent for a medical intervention (Health Care Consent Act, 1996; Provincial Advisory Committee on New Predictive Genetic Technologies, 2001; College of Physicians and Surgeons of Ontario, 2006).
“provincial policies, standards and guidelines” (Ministry of Health and Long-Term Care, 2006). I interpreted this mandate to include an examination of existing and emergent ethical issues related to the program, among them the policy and practice of consent. In my interviews I asked participants about their views on the possibility of implementing express consent for NBS in Ontario at some point in the future. In this chapter I present the descriptive and analytic/interpretive themes that I identified in my data on this issue.

Part I of this chapter outlines the descriptive thematic findings. A number of participants discussed their perspectives of express consent for NBS in Ontario as contextualized within their perceptions of the broader goals and objectives of the NBS program. The descriptive themes include two interpretations of the perceived goals and objectives of NBS in Ontario: “primary goals and objectives” and “secondary goals and objectives.” In addition, I descriptively present participant perceptions of express consent for storage of and/or research on dried newborn blood spots from the perspectives of those study participants who discussed these issues in the interviews.

In Part II of this chapter I focus on the analytic/interpretive findings on the topic of express consent. Of the participants who discussed express consent for NBS, many framed the issue as a tension between parental autonomy and NBS uptake — in so doing reinforcing the appropriateness of a public health ethics lens for this inquiry. This tension was exacerbated by perceptions of express consent for NBS as potentially overwhelming, anxiety-provoking, and logistically challenging — a potential threat not only to the goals of the NBS program, but also to the concept of consent itself. I interpreted these perceptions of express consent as a harm: express consent as potentially harmful not only to the goals and
objectives of the program, namely NBS uptake, but also potentially harmful to the goals and objectives of consent as conveyed through informed autonomous decision-making. I have distilled these tensions into three analytic/interpretive themes: “The challenge to newborn screening uptake: the perceived underlying harms of express consent”; “The challenge to autonomy: harms through the ritualization of express consent”; and “A search for balance: reconceptualizing express consent within the context of newborn screening”.

In analytic/interpretive theme one, “The Challenge to Newborn Screening Uptake: The Perceived Underlying Harms of Express Consent,” I explore three sub-themes that capture what some participants identified as among the complicating factors of express consent perceived to threaten NBS uptake and potentially cause harm to infants, parents, and society: the information that would need to be disclosed to achieve express consent; the role and influence of HCPs in an express consent process; and the logistical barriers that threaten to introduce mistakes and oversights.

I examine in analytic/interpretive theme two, “The Challenge to Autonomy: Harms through the Ritualization of Express Consent,” five sub-themes that reflect participants’ concerns as to how express consent for NBS could potentially cause harm to parent autonomy as a result of what I have labelled as the ritualization of express consent. These sub-themes include “True” express consent requires knowledge and understanding; “True” express consent cannot be coerced; The timing of express consent can impede knowledge and understanding and/or be coercive; Resources are essential to achieve express consent; and Doubts that parents really give “true” express consent in jurisdictions that require explicit parental consent for newborn screening.
The first two analytic/interpretive themes address what participants identified as the perceived harms of express consent not only to program uptake, but also to autonomy — tensions at the heart of many public health ethics dilemmas. In analytic/interpretive theme three, “A Search for Balance: Reconceptualizing Express Consent within the Context of Newborn Screening,” I present participants’ perceptions of what I have interpreted as the need to reconceptualize the practice of express consent for a public health intervention such as NBS in an effort to reach an actual balance between respecting individual rights and freedoms and achieving public health goals. This interpretation stems from the following three sub-themes: perceptions about a check box approach to consent; a simple approach to consent; and introducing a decision aid for NBS.

Collectively these descriptive and analytic/interpretive findings capture participants’ perspectives on the topic of express consent for NBS in Ontario. My inquiry strives to capture and interpret the range of participant perspectives and attitudes towards the policy and practice of express consent for NBS from endorsement to ambivalence to significant reservations. These perspectives collectively highlight the perceived tensions associated with express consent for NBS in Ontario.

**Part I: Descriptive Themes**

In Ontario, implied consent currently encompasses the newborn screen, the storage of blood spots for 19 years, and the use of stored blood spots for de-identified research (Newborn Screening Ontario, n.d.f; Bombard *et al.*, 2012). However, participants in my study differed in their opinions as to whether implied consent for the storage and research dimensions of NBS is appropriate. Their views on express consent for the newborn screen,
storage and research were often framed by their perceptions of the goals and objectives of the NBS program. I provide the range of perceptions below by presenting descriptive thematic categories that frame the goals of NBS according to primary and secondary objectives.

Descriptive Theme 1: The Primary Objectives of Newborn Screening

The primary objectives of NBS focus on the health of infants, contextualized within broader understandings of the public good. I identified three sub-themes as part of the perceived primary goals: 1) To identify and treat affected infants; 2) to identify infants with sickle cell trait; and 3) to store dried newborn blood spots for infant health purposes and quality assurance.

To identify and treat affected infants

Most participants perceived the identification and treatment of infants as the primary goal of NBS:

I just think back to the original tenets [of public health]. Where we’re doing it [NBS] for the public good, we’re doing it for treatments that can be picked up early, accurately, and that can be treated and therefore with better outcome. (#42, advisory committee)

I think that the goals of newborn screening are to … ah … identify children who are at risk of um certain medical conditions that can be picked up by screening. I think that they should be conditions that … that are treatable, that there's something that can be done for them. And so that, I mean it is a public health issue. It's … it's helping children be healthier and is helping reduce costs to the system um looking after unhealthy children, although it's not, obviously, without costs itself. (#1, advisory committee)

[W]ell, I think at it’s purist it’s to identify babies who have a rare disease that wouldn’t be picked up otherwise. But that you know, is serious and can be treated so we can prevent the serious consequences. (#20, treatment centre)
“Well, the goals of the newborn screening are to ... to ah ... for early detection of babies who might have a rare disorder that if left untreated can lead to serious or a life-threatening illness.” (#33, treatment centre)

“So it's [the goals of NBS are] early diagnosis and treatment. So what they noticed they have reduced the number of complications and they reduced the number of deaths.” (#30, advocate)

**To identify infants with sickle cell trait**

For study participants engaged in the healthcare and advocacy of individuals living with sickle cell anaemia, NBS was perceived to have two primary goals. First, to identify infants with sickle cell anaemia and second, to identify infants with sickle cell trait:

*Well, it strikes me, and I'm prepared to be told I'm wrong, but I think that it's a potentially highly effective both ... and highly effective ... it identifies both the cases and the carriers. And I guess from a societal point of view, it's pretty cheap. (Pause) And, I mean, identifying both the cases and the carriers has huge potential benefits.* (#37, HCP-advocate)

**Interviewer:** [W]hat would you say are the goals and objectives of newborn screening?

**Respondent:** (Pause). It would be twofold. First of all, it’s to identify the children with sickle cell disease. Studies have shown that if those kids are identified, and a proper procedure in their treatment followed, they have a good chance of not having the complications that sickle cell can cause. It’s a well proven fact that [inaudible 27:22] penicillin, that once a child, you know, gets the proper penicillin at the appropriate time uh, a child with sickle cell ah issues and complications that it can have down the line is dramatically reduced [...] With newborn screening, you can identify the child right at birth and you can give a set of procedures that needs to be followed to make sure that this child here gets the proper care that he or she needs. Again, you know just give him, you know, a better chance to survive. So that’s a child with sickle cell disease. *The other part of it* (participant’s voice rises slightly) is a child with sickle cell trait. Which again, you know, without the trait there can’t be the disease, although they are still very young the whole idea is knowing a child has sickle cell trait (participant slams fist on the table) [...] . (#38, advocate)
To store dried newborn blood spots for infant health purposes and quality assurance

Many participants who discussed the issues of storage of and research on dried newborn blood spots felt that storing blood spots for infant health as well as quality assurance and evaluation of the NBS program fell comfortably within the program’s primary purview:

[T]here’s certainly a reason to store samples. You know, it helps improve our testing: if you ever have a child who presents with, you know, isovaleric acidemia two years of age, and you want to go back and take a look at that child’s… if they apparently screened negative, you want to be able to pull their card and go back and reanalyze it and maybe that helps inform adjusting your cut-offs. So I think there’s huge value in keeping samples, you know, there’s probably a pretty big practical, physical, you know, storage issue with that, but I think it’s important. (#24, treatment centre)

Clearly, we would want to have a repository of blood spots so you could continue the tests to make sure your equipment is working properly, to make sure your program standards are maintained, um for research and enhancement of screening technologies. That’s all within the world of screening. (#2, advocate)

Uh I see it [storage] as part of the program um for use as quality assurance (participant says rapping on the table). Beyond that, I don't think it's their… I don't think so, like I don't think they [NBS] should then be responsible to keep them in case they're needed later for diagnostic purposes or anything. Like I think they should keep them, do the test, make sure their tests/the screen results are clear and they won't need to repeat anything, keep them to improve their tests, their screening. Beyond that, I don't think they should be responsible for keeping them for potential use later for unrelated testing or research. (#32, treatment centre)

While the storage of newborn blood spots for infant health and quality assurance purposes were considered among the primary goals of NBS, long-term storage of and research on the

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35 While many participants felt that the storage of blood spots for infant health and quality assurance was considered among the goals and objectives of NBS, not everyone thought that it should be done without express parental consent (see Descriptive Theme 2: Secondary Objectives of the Newborn Screening Program).
blood spots outside the parameters of quality assurance were largely considered to be a secondary goal of the NBS program.

*Descriptive Theme 2: Secondary Objectives of the Newborn Screening Program*

Beyond the storage of blood spots for infant health and quality assurance, participants perceived storage of and research on blood spots as distinctly secondary to the primary goal of NBS — namely identifying sick infants using the most fine-tuned processes available—and, therefore, should be separated both conceptually and in practice:

> These [storage and research] are … it’s a secondary issue, right? […] Yeah, that’s a separate … to me that has to be separate from the primary you know, the primary … (thought left unfinished). (#18, advisory committee)

> I’m sorry, it’s [research for purposes other than quality assurance is] a secondary purpose, not the primary purpose and it needs to be clear that they’re saying there’s a different … I think there’s a good case for a different set of standards around the use of that. (#2 advocate)

> Um, I’m thinking about it and maybe initially I used to think they [storage and research] … it should be part [of the NBS program], but maybe that could be a completely separate part, sort of focusing on the research aspect of it so that newborn screening is at an arm’s-length. Because in reality I’m seeing the patient, I’m not sort of looking at the newborn screen spots. So, you know, maybe having another kind of body of people who are just doing the research aspects, who are involved with the consents and those kinds of things. Because when we combine the two it becomes more murky. (#53, treatment centre)

> I don't think it's their [Newborn Screening Ontario’s] responsibility or part of their program… it's not part of their goal to have these blood spots available to aid in things like identification after ah natural disasters (participant laughs) or forensics. Like the goal of the program is to identify babies with these conditions. And once they've fulfilled

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36 While such distinctions are relatively easy to make conceptually, a number of participants felt that the application of such distinctions in practice are more difficult given perceptions of the current approach to NBS and concerns around introducing consent for NBS (see Descriptive Theme 4 as well as the Analytic/Interpretive Themes for more on this point).
that goal then I think that um...they shouldn't have to... keep them for anything else (participant laughs) because that really falls outside of their goal and ... And just parents and people have the right to know what their spots are being used for [...]. (#32, treatment centre)

Although some participants felt that certain elements of storage and research were considered secondary to the primary purpose of NBS, a number of participants appreciated the research possibilities connected to the mass storage of newborn blood spots. Moreover, research was considered critical to scientific advancements and keeping Ontario at the forefront of the field:

Well I do think that it's important for the program and... and for Ministry in partnership with the program to uh... to be at the forefront of th... the field. So if there are um any additions or any changes to the panel, I think its important for uh... this province to make sure that we stay in step with those developments. And I... I mean, I feel like I've said this a lot to you, but as a total non-expert, I don't know what those are and... and nor do most people inside government so they would rely on the newborn screening program and the expert advisory group to... to let us know when there are new developments and what those may be and how we can weigh those, weigh the consideration of... of those changes. Um but I do think that that's really uh very important for the program um in a go forward basis. (#56, advisory committee)

I would hope that people wouldn’t throw in the towel and say, ‘Oh [inaudible 2:28:57] won’t even try to use these blood spots for research.’ That would be ... well, that would say, okay we want to be a third world country in terms of not being anywhere near the frontier of new knowledge. (Pause) [Interviewer: So you’re...] That’s not a good [inaudible 2:28:17/sounds like: ‘camp’]. (#2, advocate)

However, for many of the participants who discussed the issue of storage of and research on dried newborn blood spots, they felt that pursuing such avenues within the context of NBS would require, in many circumstances, parental consent:

I don't know that there's (participant breathes out) really a clear answer, but I guess what I think is, you could consent to having the prick done and to having the information sent back to your um... your doctor so that you are getting the healthcare benefits for your child, right, without consenting to the data repository and storing the sample and all of those other things. None of that is necessary for the clinical care that you need. Right?
And so *shouldn't* the parents have a choice to *decide*. "I want to *A*) (participant gives a little laugh) just get this clinical information done for my child, to make sure that they are going to be *safe and healthy* or *B*) I wanna do that: I'm also a good citizen and I realize the benefits so (slight pause) I want that information if it *helps* the government to kind of manage programs and try to make things better for the province generally, then I want them to have the information, right? But… but the *parent* should be *educated* and should *understand*; it should be *transparent* and they should be able to make that choice. (#7, legal expert)

*The researchers* often see this in the context of a *broad, better good*. Research leads to *improvements*. *So therefore*, we should be given every opportunity to do it. And ah, *I agree with them, absolutely*. I just don't think we should be doing research without people knowing *that they're being researched*. And we shouldn't develop a *screening program* whose purpose is *research*. Its purpose is *screening, not research*. Now, if you can get some research *out of it* in an ethical way and with people knowing, *that's a real bonus*. (#31, advisory committee)

See, this is … this is where um … the decision makers have to be clear on, and that’s where the public has to make the decision. Um … you want to make sure that the parents are well informed, and consent has to be *explicit* on what it’s going to *include*, and um … you just have to … you see sometimes what happens is people don’t *trust* the system. And, okay like fine, we take it now because we want to find out about these diseases, *but*, are we going to use it for research? Are we going to make sure that if we *are*, are we going to get *consent*? You know, maybe people want options on what we can use the blood samples for … you know what I mean? Or at least let them know if they want to or not. Um…I would not want my like blood sample to be used for whatever research *years* from now, because I don’t know what that research is going to be, or if it’s going to be, for example, used against me, in terms of, for example, being hired by a certain corporation or something. Like, let’s not um … (pause) *reduce*, or eliminate the *rights* of the individuals by just *enforcing* these laws to make them *responsible* for prevention or be more responsible citizens in avoiding incidences. (#39, advocate)

Possible secondary uses of stored blood spots such as research outside the scope of quality assurance practices resulted in a range of participant perspectives from “outside the scope of NBS” to “holds significant potential but must obtain parental consent.” The descriptive theme below offers the spectrum of participants’ views towards seeking express consent for the various storage and research initiatives connected to NBS.
Descriptive Theme 3: Participant Perceptions of Consent for Storage and/or Research

This theme captures the dissonance across participant attitudes towards seeking parental consent for short- and long-term storage and de-identified research, yet a unified voice regarding obtaining parental consent for non-anonymized research.

Storage for infant health and quality assurance purposes: no consent necessary

Since storage of dried newborn blood spots for infant health and quality assurance purposes was considered part of the primary goals and objectives of NBS in Ontario, there was a general sense that explicit parental consent would not be necessary for this dimension of the program:

Well I think [...] the storage for the program itself, um … I don’t think that requires consent … I think that’s … um … not necessary because I think we aren’t getting consent for that with all other programs in medicine so I don’t see why newborn screening should be different from that regard, do you know what I mean? Any … any blood sample can be used for that in a lab, cuz I mean there’s various quality programs that are going on to make sure that labs are doing the right thing; but then those samples would be destroyed as part of that, right, like that would happen for a certain point, and then they’d be gone […]. (#3, advisory committee)

Respondent: So just due to the nature right now of the implied consent, is it like the worst thing in the world? No but I don’t think it really should be done without having a formal consent process.

Interviewer: M’hmm. For the storage as well or just the…

Respondent: No, just for using them for research purposes. I like the idea of it being stored for clinical, you know, going back for clinical presentation and practice and being able to pull out old samples to reanalyze them and go back and adjust your cut-offs and that kind of thing. (#24, treatment centre)

Although there was a belief among some participants that storage of blood spots could occur without express consent, the legal expert featured below maintains that parents should be informed that such storage occurs:
You know there are … there are … informed consent legislation for example provides exceptions to the rule for … or privacy legislation … also provides exceptions to the rule of informed consent; that, like for example for quality assurance purposes, for ensuring … for ensuring appropriate organization of the health care system, we kind of give access to our health information all the time so it's not that I think informed consent has to be given for everything, but at the same time, if it’s … if the idea is really to kind of … I don't see why they wouldn't you know why they would not tell people that blood is stored and that it may be used for future … and how it will be stored and how long is you know … with identifiers kept in place or not. And so all of these things I think will be important. (#14, legal expert)

**Consent needed or advisable for storage**

Although a number of participants felt that consent for the storage of blood spots was unnecessary, a few participants felt that parents should provide consent before the newborn blood spots are stored:

**Respondent:** Obviously, I guess, that needs consent I think.

**Interviewer:** To have it [blood spots] stored?

**Respondent:** Mm-hmm. I would think definitely.

**Interviewer:** A more explicit consent.

**Respondent:** Yeah. I would think written consent I would think for that [storage of blood spots]. Yeah. Um … yeah … If not, then I think they should be discarded, so.

(#33, treatment centre)

Seeking consent for storage and other secondary uses at the outset would, for some participants, render the secondary uses of stored blood spots less troublesome ethically:

Well, I mean that's… that's [storage and research is] certainly another argument for informed consent. It makes it much much easier to justify those kinds of secondary uses cuz, you know, people have consented for them or not. And then, you know, clearly these ones have [consented] and you can use those samples, or these ones haven’t. It gets a

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37 At the time I conducted my interviews, information about newborn blood spot storage and research was neither included in Ontario’s NBS brochure nor in the Parent Information Sheet attached to the blood spot requisition form (see Appendices F & G respectively). (The Parent Information Sheet does, however, intimate that the blood spots may be stored and used in some way, but directs interested parents to the website rather than provide information on the form.) Both the brochure and the information sheet refer parents wanting more information to Newborn Screening Ontario’s website. Parents will find answers pertaining to storage and research under the Frequently Asked Question tab of the “Parents” section on the program’s website.
little more grey when you're into mandatory or enforced consent [sic] and... I don't know, it becomes a more difficult ethical question, I think, yeah. (#12, advisory committee)

**Different opinions regarding express consent for de-identified/anonymized research**

Some participants felt that consent would definitely be necessary if any kind of research is conducted, regardless of whether the blood spots are anonymized:

I think _that_ is a real problem. You know, if you're doing research without letting people know, you know, that's concerning, and I don't think we should be doing that. I think you know, I can kind of understand where it's just, there's a temptation there, it's like, wow, you've got all these things you can test for anything under the sun. And you can, say argue that it's not, you know, if you de-link ... it's not linked to anybody ... who does it harm? But I think that's a real issue. [...] Doing research without consent is really definitely frowned upon unless there's a very real ... the risk benefit can be really justified. You know, if there is some illness taking over society and you need to ... and you needed ... and you can help sort it out by doing testing on newborn samples, I think, you know, you'd have to really be able to justify it. I think it is very unlikely, the whole thing but, certainly, no. Nowadays you have to get consent for using it even when it's anonymized. (#43, treatment centre)

But...but there's a distinction to be made between the care you can provide to the individual child who is already born and here, right (participant gives a little laugh), and what you can do for the betterment of society in the end. And I think that parents deserve the right to decide. Like we don't have a mandatory... it's your civic duty to participate in research yet, right? That isn't the framework that we operate it in? And until it becomes the framework, then I think parents deserve the right to decide how benevolent, how...how contributing to that they want to be and they can only do that if they have all the information. (#7, legal expert)

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38 I think this participant meant to say “enforced screening” rather than “enforced consent.”

39 In the interviews I took for granted a shared understanding of the language of “de-identified”, “anonymized” and “non-anonymized” to describe the various research possibilities that could be conducted on the blood spots unbeknownst to parents. While this assumption is a study limitation and future focused research should be conducted for a more comprehensive inquiry into perceptions of consent for the storage of and research on dried blood spots, these findings nevertheless reveal that some participants do not think any kind of research should proceed without parental consent.
That's [epidemiologic or clinical research is] where I think you would need more explicit consent. I think people would have to know that they're agreeing or not agreeing to that. So I think if you're going to have a mandatory model or you're going to have an implied model, then that is a—I have ethical concerns about that. Because then people don't know what their blood is being potentially used for. So someone's going to take it and start genetic sequencing on a whole bunch of babies or, you know, without people knowing. I think that's a dangerous thing. Now, if there's a … I have no issue with it if people are counselled about that and know that that's a possibility and are willing to consent to it. (Slight pause). So personally, if it were my child, I would say fine, sure, I don't have a problem with it. But I'd want to know that I knew that, right, not that I just had the blood taken and by the way, we keep it for 30 years just in case we want to do something else with it. (#31, advisory committee)

So… and then if you were to keep them later on and provide them to researchers for … for their research um … I think parents need to be aware of that possibility and I don't think, I mean I was barely aware (participant laughs) that … of how long they stored them so I don't think parents would be aware of that at all and I think you would need their consent to release the blood spots for research. (#32, treatment centre)

But we now have to fully embed in our programs that that blood spot is DNA and that is identified in the identifiable information if you have the gizmos to do that. Um (pause), so if you’re actually sharing blood spots or using blood spots, you can get [inaudible 2:20:59] it’s a falsehood to talk about de-identified, right. Statistics can be seen in a completely different light, but the actual blood spot is DNA. […] To go beyond, and I think there is a chance for asking for consent by the parents for future research ideas beyond some definition of quality improvement/quality assurance in newborn screening. It is conceivable that the DNA could be used for damn near anything, and that … and there may be perfectly justifiable examples that I … first of all in my way of doing this this [inaudible 2:22:28] society, in this case the government and public health [inaudible 2:22:27: organizers], are saying, we are making a decision in the best interest of the child. We are going to take the blood, we are going to do the hearing screen, et cetera. It’s not really up to you mom and dad. But, we now realize, unlike the hearing screen where you’re not collecting DNA, we are collecting DNA. So now we got it… and the DNA could be … it’s an incredibly valuable biobank, right. Not just for newborn screening, potentially. If you’re going to use it for a distinct person that is distinctly different from newborn screening then, I'm sorry but you gotta get consent. (#2, advocate)
Some participants, however, felt that given the public health mandate of NBS, as long as the blood spots are anonymized for research purposes, that the necessary safeguards are in place, and that Research Ethics Board approval is sought, express parental consent is not necessary:

I think the implied consent model that we have would fit with anonymized research and I think that that can be enormously useful if one is looking, as they have in Quebec, at population prevalence of disease or if one needs samples to test a new hypothesis or a new technology. Potentially even for biobanking purposes but only strict purposes that were anonymized. I am personally comfortable with that as an intrinsic part of the program. (#42, advisory committee)

Well you would need consent [for epidemiologic studies], but I don't think you'd need consent from a specific individual. So some kind of ethics board would have to review it to make sure that it was appropriate. (#34, advisory committee)

I think the anonymized use of the samples, you know, probably can have slightly more lax rules, but uh… certainly any kind of identified data would have to be very strongly linked to the overall goals of the newborn screening program to justify using that for a secondary purpose. (#12, advisory committee)

Respondent: I don’t think the issue of consent even needs to be raised personally. But that’s my personal opinion. […]

Interviewer: Does it change at all given the storage and secondary uses?

Respondent: (Participant jumps in immediately) No. No. I have no problem with using people’s data anonymously to validate new procedures when they are for a health benefit. I have no problem actually using them, even if they have some commercial extension, although I think that the value of that commercial activity should be paid back to the population, which doesn’t happen often. I have no problem ah (pause) using (pause) identifiers when the information is clinically significant. If it is likely to alter care then the identifier should be made available, because you are then in — you are aware of medically significant knowledge that could make a difference in somebody’s care, and to sit on it and not act is unethical. […] if you have a bank of 300,000 specimens from kids that were born for the last three years, then you have a new analyte that can tell you whether .02% of those kids are susceptible for some potentially fatal disease, you can go back and do that; I’d go back and do that. And if you got those positives I’d contact them and say, we have a result you need to know about. I don’t have an issue with that at all. And I think that some people would complain if you didn’t do it, and some people
would complain if you did do it, but I think that medically in a public health perspective that’s the proper thing to do. (#11, advisory committee)

Although participants had mixed views about whether express parental consent was necessary for de-identified research initiatives they were clear that parental consent was essential for non-anonymized research.

**Express consent needed for non-anonymized research**

Participants who discussed the issue of consent for research felt that express, informed, parental consent would be required for non-anonymized clinical research:  

I think that that would have to be then ah very carefully...That’s a different process entirely. And I think that for that I think you do need informed consent. If you were prospectively going to use that and, in effect, enrol somebody as a subject in an experiment, that’s different: that’s completely different. But if you’re sitting on information that you know is medically significant, then I think you can [inaudible 23:04]. *If you’re going to go looking for something* that’s different, that’s a defined experiment. You have to go through REB [inaudible 23:15] and you have to have informed consent. *That’s a completely different situation.* (#11, advisory committee)

You know, if it was research it would have to be anonymized or if it wasn’t anonymized we would have to get consent from all the parents and all of that stuff is actually in the parent’s section on [Newborn Screening Ontario’s] website. (#21, treatment centre)

I think that they [the blood spots] are an *enormously* important *resource* for the right sorts (participant bangs the desk) of *long-term research*. I think that that should either always *be anonymized or* if we’re going to use it for anything other than anonymized research, there ought to be a *clear explicit* question about whether parents want to participate in a research program *on the card*, well informed, *at the time of the test*. (#42, advisory committee)

*Clearly* if there was ever going to be any linkage or ever any results discovered as part of research that would have clinical implications for *older* babies or children, then there would, have to … I think, have to be a more active consent process. (#10, advisory committee)

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40 These views are in line with current Newborn Screening Ontario research practices.
There was a consensus among participants who discussed consent for storage and research that express parental consent was critical for non-anonymized research initiatives. However, participants disagreed on the appropriate approach to consent for de-identified/anonymized research. Some participants felt that the public health nature of NBS did not require parental consent. Others felt that while not seeking parental consent was not “the worst thing in the world”, obtaining express consent would eliminate uncertainty and doubt regarding the use of the blood spots for such secondary purposes. Some participants, however, felt that express parental consent was critical for any kind of research. These range of these perspectives – with many thinking that express consent would be preferable and/or remove some of the ambiguity around future uses – suggests that people connected and committed to NBS in Ontario are hesitant about the province’s decision to use implied consent for the storage of and research on dried newborn blood spots. A concern that was identified as an area requiring further inquiry in 2001 by the Provincial Advisory Committee on New Predictive Genetic Technologies: “The practice in Ontario of banking samples obtained from newborn screening, and the process of informed consent for banking and the future use of samples, require further review” (p.36).

Descriptive Theme 4: When Considering the Implications of the Secondary Objectives of NBS, Remember the Primary Objectives

Although many participants who discussed such secondary objectives as storage and research for NBS appreciated the possibilities afforded by the collection of dried newborn blood spots, they often returned to emphasizing the primary purpose of NBS. Specifically,
infant health should not be compromised in pursuit of secondary objectives such as storage and research or other initiatives whereby the infant does not benefit directly:

So, for me, it's definitely to prevent from the burden of the impact of having an inborn error or a genetic disease. There are some secondary reasons to do newborn screening, but this is why I said really, there is one goal that I would see as the major reason. Because the other goals are secondary and one has to see what is the balance between benefits and risks. So, I’m rather one of those with a little bit more conservative approach to newborn screening. (#36, treatment centre)

Well, the clearest and easiest and most direct benefit [of NBS] is for the infant. There's no doubt, though, that a healthy infant needs a healthy family. Right, so obviously, there are secondary benefits to the family. I think, though, if we get to the point where we use newborn screening as a means only for the family, where there's no benefit to the infant, that's not on for me. So (pause) again, getting back to the principles of screening, it has to be a condition where the early identification leads to early treatment, which improves outcome. But that outcome has to at least be rooted in the individual being screened, not just to society or to the family. So ‘it's good for me to know, but it doesn't matter for you’—that's not on for me personally. (#31, advisory committee)

According to some participants, part of protecting the primary goals of NBS meant being wary of seeking express consent for storage and research at the time of the initial heel prick. For although many participants articulated a clear need for express consent for storage and research, they also articulated their concerns about the potential implications of introducing consent for such purposes. Specifically, some participants worried that if the storage and research dimensions of NBS, were introduced to parents at the time of the initial screen and parents were confronted with the responsibility of having to make explicit decisions about the storage and research dimensions of NBS parents might decline the newborn screen altogether. (This fear is explored more extensively in Analytic/Interpretive Theme 1.) Given this fear, a few participants were adamant that if express consent were ultimately incorporated into Newborn Screening Ontario’s program to accommodate
perceived secondary uses of blood spots (e.g., research) and began to noticeably impact screening uptake negatively, they would prefer to abandon the secondary objectives altogether rather than to compromise in any way the primary goals of the program:

And for me personally, I mean if...if that [asking parents to participate in research at the time of the heel prick] were forced upon us to some extent ... where... where they... the system then was negatively impacting on what our ultimate goal is, which is to find sick kids and treat them, then I would sooner destroy the samples after a certain period of time and put a blanket ban on them. But I think that's ah ... that would be somewhat of a loss. You know. Again, it's that balance. (#15, advisory committee)

I think that you can't compromise the primary goal, which is to do the heel prick to have healthy children (participant says with a little laugh). I think that is the primary goal and anything that would compromise that goal should ... should be pretty carefully thought about. Yeah. I didn't think of that. Well that would be ... because there are some people when you see the word research they just completely want nothing to do with it so it might be the best thing for their...yeah, like I said some kind of consent. Or maybe for the ones you wanted to do prospectively or offer a consent form. (#34, advisory committee)

This fear is seemingly exacerbated by the perception that while the distinctions between primary and secondary objectives were considered relatively easy conceptually, introducing such distinctions in practice was considered more difficult. Specifically, since HCPs do not currently engage parents in conversation about whether they wish to pursue NBS for their infants, the mechanisms for supporting such interactions on the topic of storage and research do not exist:

I think it...the fact that it [storage and research] has piggybacked onto a once emergency like health screening program and... is what makes it a little complicated. It’s hard to separate it in time and place from its clinical importance...the clinical importance of newborn screening is hard to argue with for a specific group of conditions. The fact that this is automatically attached to that is what makes, in my opinion, makes it a little bit complicated. Because in other...in other situations where people are requested...where samples and information is requested of people...it's usually...it’s often separated in time and space from a clinical encounter. Or it’s...it’s a little more explicit like if you were to compare it to a tumour bank and someone going in for cancer surgery and a piece of their
tumour…their tumour has to come out and a piece of their tumour is going to a tumour bank … for the same kind of research. They know they're having surgery. There is a consent process for the surgery that is a live real exchange with a person that explains the benefits and risks. So there's a mechanism to engage someone in the research component of that. In newborn screening, that doesn't really exist, because you're not engaged in that kind of transaction for the heel prick. And…in my mind, that's what makes it a little more complicated. But how and if they can be separated? I…I…I'm doubtful. I'm skeptical. But then on the flip side I think people don't know or aren't … you know every time you go to a doctor something about your encounter is logged in a system, in the province, at ICES, it's tracked, and it’s used by health service researchers, by health systems managers to figure out how to allocate funds, to figure out budgets, to figure out systems of care…so people's information is being collected, and logged, and stored in registries all the time. And I don’t think people really know. (#27, advisory committee)

Finally, the fears and challenges connected to consent for storage and research are exacerbated further by participant misgivings about explicit, express consent within the context of NBS in general: perceptions which I explore analytically and interpretively in Part II below. For even though the heel prick, storage, and research are currently encompassed within an implied consent approach for NBS in Ontario – an approach to informed consent perceived by many participants to have its own challenges and limitations (see Chapter 6) – a number of participants shared their concerns about seeking express consent for these three components of NBS. In the second part of this chapter I frame my participants’ concerns about and ambivalence towards express consent for NBS in terms of a classic public health ethics tension: promoting population health versus protecting individual rights and freedoms.

**Part II: Analytic/Interpretive Themes**

The possibility of express consent for NBS in Ontario generated mixed reactions from many of my participants, ranging from endorsement to resistance. However, the dominant theme that I identified in my data on this issue was the perceived challenge of finding a
balance between respecting and promoting the rights of the individual through parental consent while also achieving the goals of the program, namely screening uptake:

*And then the big fear is* that if you *overemphasize* what's *concerning* and *worrysome* and value laden about newborn screening, that people are going to opt out of it altogether and then it's not gonna pick up babies with PKU. *And that would be seen as a major failure.* You know if the program suddenly is *such* a proponent of choice that it’s failing to identify sick babies, things have gone too far the other way. So how do you *retain* the importance of *choice* and the importance of *um… sort of honour* the fact that this is not straight forward, while still maintaining the primary goals of the program? It's certain, you know, certain people would...would...would *argue nothing* that even comes close to compromising the primary goal of the program should be entertained. Which is why *those* peop…people in that *camp* would say, ‘There's no place for choice here, cuz *I'm too afraid* of choice reducing uptake and resulting in babies that are sick that could have been avoided.’ That … whose sickness could have been avoided. (#27, advisory committee)

Well I think *that's* going to be the biggest driver, is really looking to see that *any process* that's put in place does not *impede* the fact that every baby should be screened. And I think you have to make, you have to realize that it's got to work in, you know, a 120,000 births all over the province in *very different* settings and so it can't be a research (the participant grabs my consent form as an example of what consent *cannot* look like and waves it in the air laughing) this kind of a consent process. It's got to be *practical*, it has to be *easy to institute* and it has to *not impede the process* but *at some level* it should help make sure parents are more aware of what's going on and make sure they understand that if they *really, really* don't want to do it, they don't have to, but what the risks are associated with declining. (#10, advisory committee)

This tension between choice and uptake captures what many public health ethics scholars have identified as the core ethical dilemma of public health (Holland, 2007). That my participants discussed this tension so directly further solidified the appropriateness of a public health ethics lens for this analysis.

The analytic/interpretive themes presented in this part of the chapter (Part II) examine participants’ perceptions of explicit informed consent as it pertains to each side of the tension — uptake and autonomy. I will explore the following three analytic/interpretive themes in

Analytic/Interpretive Theme 1. The Challenge to Newborn Screening Uptake: The Perceived Underlying Harms of Express Consent

I identified the tension between choice and screening uptake as a dominant theme in my data. At the centre of much of the discussion around express consent for NBS throughout the interviews was the underlying fear that parental choice as exercised through explicit consent could compromise screening uptake and, ultimately, the public health goals of the program. I identified three main sub-themes that capture many participants’ concerns regarding introducing express consent for NBS: 1) Information and express consent: the potential to harm infants, parents, and society; 2) HCPs and express consent: their ability to influence uptake; and 3) Logistics and express consent: the potential for mistakes and oversights. I interpreted participants’ perceptions of the perceived challenges of express consent for NBS within a broader interpretive structure of harm. Specifically, the potential for express consent to cause harm to the goals of the program: to reduce the uptake of NBS, and, by extension, harm infants, their parents, and society more broadly.

Information and express consent: the potential to harm infants, parents, and society

Many participants focused on the nature of the information that would need to be conveyed within an express consent approach to NBS as one of the main concerns introduced
by the possibility of express consent for NBS. The quantity, complexity, and nuance of the
information that would need to be disclosed to parents pertaining to the heel prick, the
number of conditions on the screening panel, storage, and research was perceived by many
participants to be a source of concern that could lead to overloading, overwhelming and/or
confusing parents, which was perceived as a threat to screening uptake and as a result a threat
to infant, parent, and societal health. In the sub-sections that follow, I present participants’
perceptions of how express consent for NBS could harm each of these three groups.

*Information and express consent: a potential harm to infants*

The participants in my study predominantly expressed their concern for infant health
— the primary demographic targeted in this public health program — and what express
consent for NBS might do to infant health. Express consent was perceived as a potential
harm to NBS uptake and infant health for three reasons. First, many participants felt that the
number of conditions on the panel, the complexity and nuance of each condition, and the
potential for ambiguous outcomes following the screening were among the reasons against
implementing express consent for NBS. Specifically, they feared that the sheer magnitude of
the information would be overwhelming for parents and/or difficult to understand. If parents
are overwhelmed or confused by the information, then participants worried that they might
decline screening, thereby leaving their unscreened and potentially affected infants
vulnerable to irreparable harm or death:

The arguments about consent are the … the *leading argument* made *against it* [express
consent] is the *fear* um that um the main purpose of newborn screening will be
compromised and that um bec… if … if parents have a lot of consent arguments to think
about, that they’ll say, ‘no, I don’t want my child to be screened,’ and then you’ll miss a
case that could have … where there could have been a difference made by therapy. So
that’s the leading argument against it. (#47, advisory committee)
I mean it would be really unusual that anyone would say ‘no’, and so then you get into … so if you present the grey disorders … um … there’s the worry that people will get preoccupied by that and not have the ones for which there’s clear evidence; so if there’s some worry that if you give true choice and true informed choice, will people be dissuaded from having it at all? Which I think many people would be concerned about, you know what I mean? So there’s kind of an um … it’s very complex of how to affect the system and not cause harm either way, I guess is the way to put it. (#3, advisory committee)

Second, some participants focused on the heel prick itself. They expressed concern that describing the basic process of the heel prick to parents might lead some to decline screening on behalf of their infant simply to spare their infant from the perceived “trauma” of the prick:

**Respondent:** […] I think most parents would still go ahead and do it. Most people don't have problems with the screening. They worry more about the blood draw. (participant laughs)

**Interviewer:** And they worry about the blood draw, just—

**Respondent:** Oh, it's going to be painful, it's going to be, you know, whatever. It's my baby. (#31, advisory committee)

It was not in the best interest of that medical professional’s child not to be screened for PKU. Not for the sake of saving a heel prick and six or eight drops of blood. I mean the risk of infection from that, is minuscule compared to the consequences of the missed case of PKU. (#2, advocate)

Third, some participants perceived the storage and research dimensions of NBS as further complicating the policy and practice of express consent. As mentioned earlier in Part I of this chapter, a number of my participants maintained that express parental consent was necessary for some – if not all – storage and research practices connected to NBS in Ontario. However, despite the insistence on consent there was an associated concern that introducing these dimensions into an express consent process could lead to added confusion for parents and/or introduce an element of mistrust or scrutiny around the language of research and potentially lead to a decline of the initial newborn screen, in so doing, leaving their
unscreened infants vulnerable. These potential reactions were among the arguments put forth against incorporating storage and research dimensions into an approach to consent for NBS:

*That’s been the concern* [that introducing issues of storage and research related to NBS would detract from the NBS itself]. I don’t know whether I have an opinion on whether it will or won’t. I think there are two aspects, one it would introduce enormous complexity into the *education*; because if that really ... if that was truly *informed consent* I think there would have to be more *education* around the heel prick test than we currently think there is (participant says with a smile). And I think it is a *real barrier*. You know, you introduce that concept and parents can *misunderstand* and feel that this is exposing their *infant* to *unwelcome identification* and *scrutiny* if they don’t really understand how it would still be *protected* and confidential. So, (a) you’ve got a lot more work, a much greater cast, and (b) you’ve got perhaps the possibility for misunderstanding and then barriers to the effectiveness of the program. And I can *absolutely* understand *all* of those and appreciate that they are *real*. (#42, advisory committee)

*Oh, yes.* If you were going to do research with those newborn screening [blood spots], you would have to get consent. So you would have to then get consent from all the people with babies, you know, so, for whatever reason you want to do research on those newborn heel pokes, they would then *have to get consent* and I think *problem* ... the reality is that *won’t happen* because otherwise ... it might ... *To do that*, then you'd have to delve into the whole newborn screening *itself* and that could really interfere with the ability of newborn screening to do its job, so I don’t think that’s real. You know, so I ... I don’t think that ... that’s *[research is] certainly not going to happen. (#43, treatment centre)

*Now we get into another* problem is that if you say that, okay, at the time that you consent the person ... or the screening test is *done* – say in the nursery – you ask the parents – the mother usually – ‘Would you consent having this sample *stored* and access to it be made available on the basis of informed consent in the future for whatever purposes?’ The very *fact* that the sample might become *accessible* in the *future* was thought to be a sufficient *dissuasive* to participate in screening *in the first place*, that people might opt out of having their babies tested. *The best way to ensure* that nobody can access your baby's test is not to have the test done. And ... and since we have no *feeling* about the ... the uh *intensity* of the feeling of people about this and ... and we played around for a long time with the wording of a consent that would ... that would *allow us* to store the samples uh for the purposes I *described* [infant health, methods development, and surveillance] and *not* put people off the screening process altogether but uh that’s ... that's a work in progress. We still haven’t resolved that. (#54, advisory committee)
The perception that parents could potentially decline screening on behalf of their infants due to the multi-dimensionality, complexity and overwhelming nature of express consent was a factor that, if realised, would be perceived as “impeding” or “interfering” with the goals of NBS. As such, parent decisions not to have their babies screened would leave infants vulnerable to undiagnosed disorders that could lead to irreversible infant harm, including death.

*Information and express consent: a potential harm to parents*

Although the possibility of express consent causing infant harm was the predominant concern, I also identified two additional groups of people whom my participants perceived to be “at risk” if an express consent policy and practice were implemented for NBS: parents and society as a whole. With respect to parents, some participants expressed concern that the process of express consent could potentially increase parental anxiety unnecessarily—which I have interpreted as a perceived harm. Some participants felt that in the grand scheme of parental worries around pregnancy, childbirth, and delivery, NBS should not necessarily be on the top of those lists given how few infants are ultimately affected by one of the conditions on the panel:

So um … a lot of people would argue, ‘what’s the need to talk about it when the odds are the person will never have a baby with that, why worry them, why upset them and involve them in that when the odds are really low that they’re going to have a baby with one of these disorders.’ So…so it’s a challenge how to figure that out. (#3, advisory committee)

[W]e all are really concerned about newborn screening and think it’s *really really important*. But in the context of getting your car seat, and having a baby and giving birth (participant says half smiling/laughing) and … and, you know the kids at home and ah HIV screening and ultrasounds, and you know *all the other things that are worries*, newborn screening for things that are going to end up being 1 in 700 as a possibility of
being real, is not foremost on people’s minds: and perhaps neither should it be. (#18, advisory committee)

So, that’s the ... and, I mean, the pregnancy and all the preparations is already scary enough, so they learn about so many risks that child is having that you can forget that it’s still a normal thing to get a baby; yeah. And increasing this by (participant starts laughing) extended newborn screening information is perhaps not the right way. (#36, treatment centre)

Not all participants, however, felt that express consent would increase parental anxiety.

Rather, a few participants were clear that in their professional experience working with and caring for parents whose infants had screened positive for a condition, parents who were familiar with NBS were perceived to be less anxious than parents who were not familiar with the program:

**Interviewer:** And in your view or opinion, do you have a sense of what might help reduce parental anxiety?

**Respondent:** Um, Good… well two things, one, good informed consent. There is a huge difference between someone who knows (participant gives a little laugh) that a test was done and someone who doesn’t. You’re starting off from an entirely different perspective, so that’s one like huge piece I think. And the second one is like appropriate staff and resources to follow things up. (#24, treatment centre)

Another sub-theme that I identified in my data that supports the larger interpretation of express consent as a potential harm to parents, is the perception of express consent as a privileging device, raising questions about social justice. Specifically, some participants articulated their concern that some parents in Ontario would be disadvantaged by the implementation of express consent for NBS. Two issues in particular were discussed:

challenges to the timing of obtaining consent and the comprehensive and complex nature of the information that would be part of that consent.
Challenges to the timing of express consent pertains to the ongoing debate about when express consent for NBS would be sought in the event such an approach were implemented in Ontario. There was a general sense that obtaining consent in the prenatal phase was preferable to consent following labour and delivery (see Analytic/Interpretive Theme 2 below for more on this). However, there was a perception that not everyone seeks prenatal care or attends prenatal classes:

[Y]ou know they say, ‘Well, what about prenatal classes?’ Great, great idea. Not everybody goes. (#27, advisory committee)

Some participants were particularly concerned for lower-educated and lower-income women, as there was a perception that more socially disadvantaged women are less likely to seek and/or have access to prenatal care or prenatal courses:

But then, can you force parents to go to the prenatal courses? (Short pause) Probably not. And again, I think that that will reflect those people who are going to the course or the session, I’m not sure what’s the name now. Those, I suspect, have education that are high school or above, [inaudible 32:04] because to be able and to decide that you want to go to the courses you need to be able to understand that you will be sitting for two hours and you will just get … receive information. And there are some people that are not interested in that. They just want to be done with the pregnancy and move on. (#17, advisory committee)

But, so … it’s very clear that they don’t … they do not seek health care as well – the poor people […] So, it’s a problem with poverty (participant claps hands together softly). (#50, treatment centre)

If express consent is ultimately tied to prenatal care, some participants feared that parents in lower socioeconomic positions may not access prenatal services and therefore the opportunity to consent would be missed and infants with rare disorders could be missed,
compromising the goals of the NBS program and the burden of care disproportionately affecting certain members of society.41

The second concern connecting express consent to issues of social justice pertains to a criticism of express consent more generally (see Chapters 3 and 8): consent forms are perceived as often challenging to read, digest, and comprehend. As a result, some participants were concerned that individuals with lower literacy and education would be particularly disadvantaged and further compromised by an express consent process. This concern was exacerbated by the perception that disseminating information about the NBS program at an appropriate and accessible reading level is challenging: “So it’s [information about NBS is] on the [Newborn Screening Ontario] website and hopefully there to help people out, it’s just difficult to convey at a grade 4 level” (#21, treatment centre).

The current approach to NBS carrier status disclosure for sickle cell anaemia that was implemented in Ontario on November 1, 2010 was mentioned by a few participants as an example of an approach to express consent that was perceived to introduce the potential for creating distinctions among certain members of the community — those who are aware that they have the option to learn their infant’s carrier status and those who remain unaware. More specifically, Newborn Screening Ontario offers parents an opportunity to learn whether their infant is a carrier of the sickle cell trait — a diagnosis made possible through NBS technology. Parents decide, with their HCP (or with a HCP connected to Newborn Screening Ontario if they do not have a doctor), whether they want to learn their infant’s carrier status results. However, this approach was perceived by some participants as potentially

41 This concern also has implications for the perception that express consent may not foster autonomous decision-making, the focus of analytic/interpretive theme 2.
privileging those individuals who first know what carrier status means and that such results are available through NBS:

The plan, right now, is that people need to actively contact the newborn screening program to find out if they are a carrier. So basically, it’s up to the … now it’s up to the … the plan is that it’s up to the people out there to say, “I want to know if I’m a sickle cell carrier.” So they need to know (a) what the heck is a sickle cell carrier. You know, your average person knows nothing about it. They don’t know … and how would they … it’s just going to be one line on an information sheet that they get, and so they’re going to have to talk to their family doctor and then their family doctor is then going to have to contact the newborn screening to find out the results, so it’s a very, sort of, active process. They have to actively seek it out, which I think is going to be very ineffective. That’s my opinion, anyway. (#43, treatment centre)

In addition, some participants felt that the current approach to consent for carrier status results would most likely benefit individuals who would have actively sought carrier status testing independently from NBS. A few participants maintained that parents who remain uninformed about NBS and sickle cell carrier status will likely remain uninformed about the availability of accessing such information through NBS, thereby challenging the extent to which they ever had a true choice:

[I]t seems to me that if it’s [consent’s] a process where it’s very passive, all that’s going to do is … it’s going to create the people who would get tested through another route get the information without getting tested. Uh, whereas the people who don’t have any information or knowledge about it aren’t going to … wouldn’t get tested later and they’re not going to use this information that they may consider valuable is again, going to be lost to them. There’s no way that they will have knowledge of it or other things, that it’s possible. Whereas if you have a family and they have members affected by sickle cell disease, they’re going to find out that information, but they would have got the kids tested at some point as they were growing up anyway. Um, so I probably wouldn’t be in favour of it for that reason. I just … I don’t think it has a lot … you’re not going to get really any increased … the only benefit from it is it’s going to save some testing down the line in terms of cost, but it’s not actually going to benefit more people, because I think the people who are going to seek out that information would seek it out at a later point as well. (#45, HCP-advocate)
This theme, “The Challenge to Newborn Screening Uptake: The Perceived Underlying Harms of Express Consent” predominantly reflects the perceived challenges express consent can pose to NBS uptake. This sub-theme explored what some participants identified as the potential for express consent to cause harm to parents, particularly regarding the possibility of unnecessarily increasing parental anxieties for conditions their infants almost certainly will not have. (Although, it is important to remember that some participants were adamant in their belief that informing parents would have the opposite effect and actually decrease parental anxieties, particularly among parents whose infants screen positive.) Other possible harms to parents articulated by my participants focused on the influences of the social determinants of health. For instance, if express consent alienates certain individuals and consequently negatively impacts NBS uptake, the potential implications from refusing NBS (e.g., unscreened affected infants) may disproportionately affect certain members of a population. However, in this example, it is easy to see how overlap exists between the two overarching themes of consent causing harm to both screening uptake and parent autonomy (see analytic/interpretive theme 2). Parents who may decline NBS for their infants based on a lack of knowledge and understanding and/or feeling burdened and overwhelmed, raises questions about the extent to which the approach to consent implemented succeeds in supporting and fostering informed choices for all parents in a community, a theme explored in greater detail later in this chapter (see analytic theme 2). The final theme that I explore in the next section of this overarching category of “Information and express consent: a potential to harm infants, parents, and society” is participants’ perceptions that express consent could cause harm to society as well.
Another theme that I identified in my data addressing the perceived tension between protecting parental autonomy and NBS uptake if express consent is implemented for NBS in Ontario, was the perceived potential harm to society at large. If introducing express consent were to adversely affect screening uptake, there was a sense among some participants that the broader societal, common good would suffer as well not only financially, but emotionally and morally. Many participants explicitly referred to NBS as a benefit to society and that express consent for NBS could threaten society’s financial, emotional, and moral health.

The possibility of financial harm to society as a result of express consent was defined in two distinct ways. First, the financial burden was interpreted in relation to the costs of implementing an express consent process for NBS, costs perceived to have implications for the consent process itself as well as broader healthcare implications. The costs of implementing express consent were perceived as financially prohibitive, as some participants focused particularly on costs related to the investment in human resources to accommodate a province wide consent process. Such expenditures would, in the minds of some participants, “paralyze the system” (#31, advisory committee), compromise the consent process, and ultimately result in reduced screening uptake:

But, but then you have the implementation: we sometimes forget that… you may decide, for example, as a the… the example I gave you before with getting informed consent should be done (pause), but then you think, ‘Oh my god, that means we're going to have to hire ten percent more nurses.’ That’s… that’s a… has a HUGE budgetary implications. And in a… a… in a…. a zero sum game, that means it has to come from somewhere else. (#54, advisory committee)
So if you're going to screen, if you're really going to screen, you want to get as many as you can. And if you had to rely on a big, long conversation with everybody, I think it would mean a lower rate—not because people would refuse, but because there wouldn't be the resources to do it. (#31, advisory committee)

Second, potential financial harms to society were enumerated within the context of unscreened infants ultimately affected with one of the disorders on the panel and, as a result, subjected to irreparable life-long harm or death. “Society” was identified by a number of participants as having the financial responsibility (e.g., healthcare costs) of caring for a sick infant whose illness could have been prevented or mitigated if identified and treated early:

Now I think actually there’s a moral imperative on society, because, under a universal health care system, we all will be paying big time for the treatments of preventable damage in neonates (participant says deliberately spacing out each word). (Pause). [Inaudible 1:54:33] as taxpayers we will have a huge burden if we ah have a policy that allows the innocent neonate to be missed. (#2 advocate)

But I, as a government, that eventually has to be the one responsible for taking care of that child. Don’t I have a right also to say [inaudible 45:22: sounds like ‘I want to know?’] Again, I think a government better know because we as a public, I don’t think we’re going to be able to stand for that, to say okay let some people decide oh they don’t want to know, when down the line, it’s the public fund that will end up taking care of this child here who could have done things differently had we known. I do not think the public would stand … but I mean, as I said, you know, I … I have never even heard or read of a … of parents objecting to finding out that their child has a … a birth defect. (#38, advocate)

The economic responsibility was sometimes discussed in relation to the larger emotional and moral responsibility of society to ensure that such preventable health outcomes are in fact prevented:

I see it [mandatory screening] as an option, not a decision. I see it as an option. And it might … in … one of the advantages is, you know, I think there is a bit of a philosophical position that if your baby has, you know, the worst case scenario, ah a woman decides to not have her baby screened and the baby turns out to have a treatable disease and by not

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42 Although participants spoke quite generally in terms of societal costs, it is important to remember that these costs arguably extend beyond healthcare costs to include education costs and judicial system costs as well.
treating results in long-term disability to the child then [inaudible 1:01:30/sounds like ‘that’s where’] society picks up the costs. You know, then we make some analogies to immunizations. And so we do have rules about, if you want to go to school, you need immunizations, so. So there are precedents, you know, in society where I think the … if there’s a distaste for doing … making things mandatory there’s also ah … an argument that society has a role to present its you know … in this decision process, because the society is the one who carries the … at least the financial and probably some element of the emotional burden for this decision then we … (participant’s voice trails off) so yeah. So … so … it’s … that’s an input into the discussion as well. (#49, advisory committee)

The participant quoted below elaborates on the perception of a perceived moral imperative that all infants in Ontario be screened for the expanded panel of conditions:

‘Okay, what is the impact of what we are doing or not doing and how is it affecting real people?’ And in this case [the case of NBS] I think that got lost. Even when they [public servants] were looking at the figures, being told 25 kids were…were dying every year. Twenty-five becoming severely disabled and uh… when you're talking money… what… what seemed to get lost too in the equation was the cost of supporting a severely disabled child in an institutional setting for the rest of their lives at government expense. It's expensive too…(participant laughs) You know it’s… and this… you know forget the moral cost. I think there is a moral and ethical cost to a society that allows that to happen and… and some of the medical advisors were starting to talk about that as well. It is not just a … a question of financial responsibility: medical responsibilities and ethical responsibility. (#55, advocate)

I asked this participant to expand on his/her interpretation of moral costs to society:

**Interviewer:** You did mention a number of times that there was sort of um moral costs to a society for not … for not expanding newborn screening and I was wondering whether you could expand a little bit on what those costs to society are?

**Respondent:** Well, I guess, uh… just generally speaking… if… (pause) if we're losing our children – and I find 50 a year to me is not something easily dismissed that we're losing – and … I think we're losing a child if … if the child becomes severely disabled and … and developmentally delayed. Um, (pause) if we're losing them and we could do something to prevent it and the costs of doing it are not prohibitive, then I think we have a moral obligation to do it as a society. If we dismiss that kind of thing out of hand… if we… if we don't pay it the attention that it deserves, I think we diminish ourselves as a society. Uh… that's… that's what I mean by it (participant says with a smile). I mean this is like my… my personal view. I think it’s certainly supported by the report [referring to the Ombudsman Report, *The Right To Be Impatient* (Marin, 2005)] and I cert… I believe to a certain degree it was the view being expressed by all those medical advisors and specialists. When they talk about the ethical obligation to do something, it
reflects that, as a society, we want to be a society that says, you know, if we can do something, we should do something. (#55, advocate)

NBS was clearly articulated as a societal benefit — with a predominant focus on the health of its infant members. If, however, the province were to introduce an express consent policy for NBS and it began to negatively affect the uptake of NBS, there was a sense that society would bear not only the financial responsibility of the potential implications, but the emotional and moral responsibility as well. I have interpreted the focus on the emotional and moral health of a community or society as part of a robust, holistic interpretation of the common good.

Within the overarching thematic category of express consent as a challenge to NBS uptake, participants discussed their views on their perceptions of HCP involvement in NBS generally as well as their potential influences within a hypothetical express consent context in particular.

*Health care providers and express consent: their ability to influence uptake*

Another major theme in my data around the tension between parental autonomy and NBS uptake was the perceived role of HCPs in informing parents and obtaining express consent. As much as participants feared parental reactions to an express consent process, they were almost equally wary of the role that HCPs would play. HCPs are responsible for informing parents about NBS and promoting parental autonomy and self-determination through informed decision making. However, a number of participants felt that many HCPs lack the necessary knowledge to inform parents about NBS and/or may have particular biases towards NBS, both of which were considered potential causes for concern within an express
consent context specifically and for the effectiveness of the NBS program more broadly. In addition, HCPs were perceived to have, for better or worse, the ability to influence parents’ decision-making and, therefore, have a direct impact on the uptake of NBS and, consequently, infant health.

Knowledgeable health care providers are essential for newborn screening programs generally and within the context of an express consent approach to newborn screening specifically

Participants emphasized that HCPs are essential to an express consent process and, therefore, it is imperative that HCPs are capable of informing parents:

So my… my personal view on it [consent for NBS] is that um… informed consent would probably be a better way to go. Um… I understand that there is some logistical problems to that in terms of making sure that the people who are collecting the informed consent are truly in a position to inform, I think is the largest hurdle. Um… I don't think that, from a parental point of view, it [express consent] would significantly impede the uptake into the program. I think others in other jurisdictions who have informed consent find they have a… still a high uptake. So it's more of a logistical problem, you know, i.e. training the nurses or whatever to truly give informed consent. (#12, advisory committee)

However, a number of participants maintained that HCPs currently are not sufficiently equipped to obtain such consent.43

[Y]ou have a lot of health care providers now who couldn’t give a full and proper consent to … to newborn screening. (#1, advisory committee)

The other model at the other extreme would be giving everyone fully informed choice um … and I just think it’s um … I’m not sure that that’s possible …that’s … that’s something I’ve been struggling with how to figure out too … cuz it’s ah […] most providers in primary care and in obstetrics, in basic obstetrics, won’t know what those disorders are,

43 A number of participants relayed what they perceived as HCPs lacking in NBS knowledge, which was considered a current impediment to an express consent approach to NBS. This perception is arguably worthy of empirical follow-up given that HCPs unable to educate parents about NBS arguably has implications for the current implied consent approach to NBS as well. For while consent is implied in this model, “consent” must, nevertheless, be informed (Health Care Consent Act, 1996; College of Physicians and Surgeons of Ontario, 2006) and HCPs are arguably integral to the success of such an approach as well.
won't understand what they are, wouldn't know the grey zones of those disorders, and so again it would come down to, you know, here is something that might help your baby, do you want to have it? And what mother’s going to say “no” to that? (#3, advisory committee)

These attitudes appear to stem from some participants’ perceptions that many HCPs currently remain insufficiently informed about expanded NBS, the conditions on the panel, and the process of screening and follow-up testing in the event of a positive screen despite targeted HCP education initiatives undertaken by Newborn Screening Ontario and the NBS treatment centres to date. All of these issues were considered to effect not only an express consent process, but also the effectiveness of the province’s screening program.

Independent from an express consent approach to NBS in Ontario, specific efforts were made to educate HCPs on the expanded NBS program through onsite visits, tutorials, and a comprehensive website. Participants shared with me many of the preliminary education efforts undertaken when the NBS program first expanded, including “correcting problems in the system where hospitals thought they were doing it [NBS] right but they were doing it wrong” (#15, advisory committee). Newborn Screening Ontario also conducted training sessions which subsequently evolved into “travelling workshops” (#21, treatment centre), whereby a Newborn Screening Ontario team of HCPs would travel to the various hospitals responsible for conducting the heel pricks to continue submitters’ expanded NBS education:

And so we have what we call submitters workshops. We started in Ottawa and we invited lab techs, nurses, midwives, and any submitters, which is a broad term from our perspective, um… to this workshop. And we talked about, you know, wha… what we do, why we do it, um… what they can do to make it easier, like fill out the data collection on the form (participant laughs), those type of things um… and how to do blood spots, you know, what are… we… when they were here in Ottawa we took them on a tour of the lab,
but then we also took these submitter workshops and we went to Kingston, we went to London and Hamilton, and did the same thing at each of the treatment centre locations. (#19, treatment centre)

A submitter workshop, so that’s like our travelling workshop […] So those are educational workshops we do for people again on the kind of ground level in newborn screening. (#21, treatment centre)

Newborn Screening Ontario has also developed a comprehensive website on the province’s NBS program with a section devoted to practitioners. Also, in addition to “paper pamphlets […] We’ve done video shows … we host a bulletin now to our submitters” (#15, advisory committee) and a “submitter’s handbook which would be like a newborn screening manual for everybody who is on the ground taking newborn screening samples” (#21, treatment centre) is forthcoming. These initiatives were designed to equip HCPs with the necessary information to implement NBS accurately and effectively. Even NBS treatment centre personnel discussed contributing to HCP education efforts by hosting their own workshops:

We’ve done some outreach with our nursery as well as the NICU at [one of the local hospitals] but I think that those would probably still be – cuz there are some nurses still, to this day, that refer to it [NBS] as the PKU test and we’ve already done (participant laughs) education outreach. (#52, treatment centre)

Health care providers perceived to lack up-to-date newborn screening knowledge despite education efforts

Despite these sustained education initiatives participants remained somewhat disheartened that many HCPs do not appear to have either learned the new information regarding NBS in Ontario or have not yet incorporated this NBS knowledge into their day-to-day regime:
I guess if I had a wish it would be nice (participant says with a little laugh)… I feel like our submitters … so the people who are taking the samples and sending them to us which is a huge population right? Um, I feel like as much as we try to um educate them um, that sometimes that message is not getting clearly across to them, so it would be nice to feel like we were doing a better job than we are, so that we could do a better job. (#20, treatment centre)

Well I think it [more HCP education] would influence it [uptake of NBS] positively if we had more opportunities to reach more people through education, because I think once people appreciate what you’re really doing, then there might be more of a stronger uptake. But (participant lets out a sigh), I just feel that no matter how much [HCP] education we’ve done, I’m not sure we’re actually reaching the people that we want to reach. (Pause). Otherwise I just can’t explain why there’s such a resistance to learning the new information [about the expanded NBS program]. Cuz, I mean, people go into health care because they genuinely care about people (participant laughs). So, it … it kind of doesn’t add up to me why there would be such resistance other than they just don’t have the information. (#52, treatment centre)

In addition to not incorporating this new knowledge into daily practice, some participants maintain, based on their professional interactions with and observations of non-Newborn Screening Ontario-HCPs, that some HCPs seem to be unaware that the program has expanded at all:

Quite a few of them [the HCPs] didn’t even realize Ontario’s NBS program had been expanded. (#46, advocate)

[T]hey [the family physicians] really had no clue about what was going on. This is sort of, just a few months back. (#53, treatment centre/interview October 2010)

[…] the submitters and some of the people who are sending in these samples should have a better idea of why they are doing the test. (#15, advisory committee)

Such opinions were formed in part by participants relaying their experiences with HCPs who either seem not to realize that NBS screens for PKU or do not realize that NBS now screens for more than PKU:
Many of them didn’t even realize that the newborn screening program was actually for phenylketonuria, which was quite surprising to me. (#46, advocate)

Other participants shared that despite the expansion of Ontario’s NBS program, a number of HCPs that they encounter within the daily practice of NBS continue to refer to NBS as the “PKU test.” Other participants described their perception that some HCPs thought NBS was only for PKU given that HCPs repeatedly refer to NBS as the “PKU test”:

Even health care providers: nurses that I talk to and hospitals, they’ll often say, ‘Oh,’ … think that PKU and newborn screening is interchangeable. (Pause). (#51, treatment centre)

The fact that many blood spot specimen cards are returned to the NBS lab with the parent information sheets still attached (see Appendix G) was considered further “proof” that many HCPs have not yet fully incorporated the program changes:

So they [HCPs] would give this [the Parent Information Letter on the top of the bloodspot sample card or requisition form] to the parents and then they [HCPs] would fill out this information and then it is carbon copied here and they take the blood on these dots and then send this to the lab. Unfortunately, we get a lot with this still attached (participant says referring to the Parent Information Letter) so we know that not everybody is using it but there is only so much we can do. We can’t force people, we can tell them what they should do and … it’s like recommendations, right. (#21, treatment centre)

Uninformed health care providers: a potential harm not only to an express consent process, but also for the uptake of screening

In addition to participant descriptions of the NBS program’s education efforts and what they perceived as HCP-resistance to the expanded programs’ initiatives, some participants also offered their interpretation of what such resistance means within the context of express consent for NBS. For instance, referring to NBS as the “PKU test” despite the significant number of conditions currently on the screening panel was considered a harm to parents, particularly as it perpetuates inaccurate NBS information. Such perceived
misconceptions among non-Newborn Screening Ontario HCPs were considered worrisome and a threat to an express consent approach to NBS:

I feel like there's a lot of… the nurses in a lot of hospitals still call it the PKU test. So we'll get calls saying, 'Oh, he's got … he's screened … he has a … a CF screen positive on the PKU' (participant laughs). You know, it just makes no sense, whatever. So I think that misunderstanding or that um … inability to change a … to realize that we've expanded could leave people at risk of not getting the right information or mishandling the sample. (#19, treatment centre)

But I think it’s kind of … it implicates the situation more if you’re telling them that this … you’re giving them information but you’re saying, ‘this is just for PKU,’ then I think it’s, you know, you’re not giving all the right information, so they may be consenting but if you had told them it’s an expanded panel, would they have then come out and have said, ‘no I don’t want this’ (participant laughs). (#52, treatment centre)

In addition, participants expressed a general concern regarding the kind of NBS information HCPs give parents:

So, I think (participant coughs) … I think people are comfortable with what they’ve always been doing, and I think that on some level there’s a resistance to change. Um, and I think there is, on some level, an awareness that newborn screening now has changed but I don’t think that that’s actually translated in um like how people are dealing with patients and like the information they’re giving them and how they’re giving it to them. (#52, treatment centre)

Associated fears tied to the perceived lack of awareness among HCPs included concerns that within an express consent context HCPs with limited knowledge and understanding could convey personal biases to parents that would not only effect parental autonomy, but could compromise infant health:

I mean, you’re going to have some nurses that, ‘Oh yeah, if you don’t want to do it, you don’t have to.’ Like, who knows what they’re going to say, right (participant says with a laugh). And maybe I should give people more of a benefit of the doubt, but I guess I worry that patients are gonna decline and then something’s going to happen to their baby, and the whole point of having these um newborn screens is because they may be rare conditions, but there’s something we can do about it. And, you know, ultimately the goal is to save babies and I think that we are doing that. (#52, treatment centre)
Respondent: … And you know, direct them [parents] to where there’s information and whether you have a handout of the benefits of it. I mean, again, I guess it’s depending on who’s delivering the message because … because they [HCPs] could have a bias on whether or not to do something; so you’re hoping that people are presenting it in an unbiased fashion.

Interviewer: So in terms of like how they’re presenting newborn screening can affect—

Respondent: Um hm. I think so, because I think the way doctors present some of the… the—and again it’s my personal perspective and it’s just more from experience in other people in terms of doing screening too. I think doctors, sometimes … their bias comes in.

Interviewer: Okay. Can you explain a little bit about some of what that bias might be?

Respondent: Whether or not to do certain screening and what your choice should be once you have that screening (participant laughs). (#48, advocate)

[I]n some areas where we have home births, this is done by midwives, in these groups we only have 95% participation rate because some of the midwives are not convinced and they see it as a severe um (pause) procedure … or a severe maltreatment to do an incision in the child. And if you see then there’s a child with PKU diagnosed because of severe mental retardation at the age of the two years that has this disease because the midwife counselled the family not to do this incision, and then it’s too late to treat this child and then you would get a different view of this. (#36, treatment centre)

In short, within the overarching thematic category of express consent as a challenge to NBS uptake, a number of participants perceived the role of HCPs as key contributors to either the success or failure of the expanded NBS program generally and an express consent initiative in particular.

Logistics and express consent: the potential for mistakes and oversights

The final theme I identified in my data that illustrates the perceived potential for an express consent process to cause harm to infants, parents, and society were the logistical

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44 It is important to note that while this participant highlights midwives as contributing to low uptake, other participants have identified midwives as more likely to give appropriate information and not let any baby fall through the cracks: “Um I think newborn screening falls between the cracks of pre- and postnatal care. And… so the prenatal care providers, the OBs, the midwives, I mean midwives actually span the spectrum which is what's really nice about midwifery, is that you don't fall between the cracks with a midwife because their...that's exactly the period of time that they are responsible for.” (#27, advisory committee)
dimensions of express consent. At a very functional and pragmatic level participants perceived a number of logistical challenges that could potentially be introduced if an express consent process were implemented for NBS in Ontario. Such logistical obstacles were identified as barriers to screening and a potential threat to infant health:

But I think there'd probably be more barriers to getting newborn screening completed with an explicit consent process—having to get signatures and present the paperwork and all those sort of things. I certainly see that being more of a barrier [...] I'm just sort of thinking about the process and the steps. You know, usually the turnaround times and to help baby in and out of the hospital and trying to get that information conveyed to the family, having them sign paperwork, be able to do the test before the family leaves and making sure it's not missed and trying to track them down later and making sure it's done in the window that newborn screening requires the testing done. I just see that being more of a challenge if it was made an explicit, you know, sign a form consent process, although definitely the parents would be more informed about what's going on in the testing. (#13, treatment centre)

Much of the logistical challenges enumerated by participants were predicated on the assumption that informed, express consent for NBS would need to be written, complete with signature:

So the decision to say insist on… on a informed, written, signed consent to screening ah in the first instance had major resource implications [...]. (#54, advisory committee)

I’m in labour don’t take any paper to me to sign: I can’t do it. (#35, HCP-advocate)

Participants trained as genetic counsellors, however, were seemingly less likely to assume that express consent would require a signature:

Yeah, and I think, you know, it doesn’t … you don’t need a signature for it to be informed consent. Like, I don’t actually envision anything being signed, I don’t think it needs to be a terribly formal process, but you can still get … I mean when I order genetic testing, you know, through, you know, for patients I see for genetic counselling, I don’t often get a written consent, but I’m still getting informed consent through discussion, and I think that’s all it needs to be. Yeah, and I don’t… and I think that’s different than implied consent. (#24, treatment centre)
Although a signed, written consent does not ensure that parents have made an informed decision, the signature dimension of written express consent was considered by some to be a safeguard:

Although consent can be written or unwritten, I mean basically … and again people often confuse the fact that there is a written consent with the fact that there is consent: you can have a written consent which just is … is not … is not good and does not provide adequate consent. So, but written consent is often seen as kind of an extra procedure to make sure that people have the time to kind of take something home and think about it and that they kind of can read through it a bit more carefully and … and it certainly ah a beginning of proof, which is quite strong. So if you have a written consent … that’s why we have written consent then … many surgical procedures it … it is because people kind of are worried that people would afterwards say that they didn't consent. Now, signature is not necessarily a sign of full consent — full and informed consent — but at the least it's kind of a … so it adds a layer of … or it's kind of an additional guarantee … or an additional … it's not a guarantee … an additional ahhh… safeguard. (#14, legal expert)

Among participants who framed the challenges of express consent within the context of a signed document, logistical challenges connected to such a process focused primarily on tracking issues. Participants expressed difficulty conceptualizing how to operationalize such a consent process given the sheer number of babies screened each year; the diversity of process across birthing venues in Ontario; the lack of uniformity regarding consent for NBS across jurisdictions nationally and internationally (which was perceived to have potentially negative implications for visitors and the newly relocated who deliver in Ontario); and the question of what to do with the signed consent or dissent form once obtained. Most of these questions, debated in the literature and among my participants, are rooted in unresolved issues as to when informed consent would be obtained — a logistical hurdle that has yet to be resolved.

Participants felt the prenatal period was the most appropriate time to seek parental consent for NBS, yet such an approach was not without limitations given perceptions that
there is great variation in terms of parents accessing prenatal care. Tracking issues related to express consent were also identified as potentially problematic given the lack of NBS uniformity across jurisdictions. Specifically, that pregnant women cross borders through relocation or travel and may give birth in Ontario was perceived as an additional challenge to an express consent process, particularly if it is sought at the prenatal phase:

[A]nd if you moved … if you’re pregnant and you’re living in Winnipeg and then you moved to London and … now you’re relocated and due in 6-weeks time, well, you know, it’s … it’s just complexity upon complexity, upon complexity. So I … I just don’t understand what the … I understand theoretically what the purpose of the consent process is, but I don’t understand (short pause) in actuality (participant claps hands) what the purpose of it is. (#50, treatment centre)

These concerns around express consent from a logistical perspective are ultimately rooted in the fear that through the introduction of express consent a mistake might be made and infants who would otherwise be screened go unscreened. Arguably connected to the logistical challenges of express consent for NBS and the variation of NBS practice and consent across jurisdictions is the perceived lack of knowledge and understanding of NBS in Ontario (which I explored at length in Chapter 6: Perceptions of Implied Consent for Newborn Screening in Ontario). One might imagine, for example, that jurisdictional differences and other perceived obstacles such as parents not seeking and/or not accessing (or not having access to) prenatal services could be easily addressed by either asking pregnant women upon presenting at the hospital for labour and delivery or prior to the heel prick whether they have heard about NBS or had a discussion with a HCP about whether they would like to have their infant screened and then proceed accordingly from there. However, the possibility exists that parents may confuse prenatal screening and NBS, which could interfere with program uptake and lead to infants not being screened.
This possibility for prenatal/neonatal screening confusion was introduced in my study in two ways. First, one participant explained his/her involvement in recruiting participants for a survey study involving women whose infants screened negative for the conditions on the NBS panel, yet, women’s survey responses indicated that they had screened positive, which my participant indicated was a sign that they had perhaps confused it with prenatal screening:

But it’s um interesting information that came from it [the study]. Just anecdotally, we got a lot of the surveys back, and they would say that they were screen positive (pause) and, considering I pulled the data myself for the cohort (participant laughs) it was a random sample, you know. But I knew that I had excluded all the screen positives so we would always go back and check um… you know, well it was… we’d look up the baby and make sure that maybe something got through or whatever. Um… and… it… there was no screen positives in our cohort, but there’s a confusion between prenatal (slight pause) testing and coming up as a screen positive in prenatal testing or at a high risk of something in prenatal testing, and newborn screening, right? (#19, treatment centre)

The second way in which this possibility for confusion was revealed in my data was through the unanticipated finding that three participants in my study – participants perceived by other participants to have played a key role in debates, advocacy, and guidance in various capacities and various degrees at the time of the program’s expansion – appeared to confuse NBS with prenatal screening:

Yeah, the newborn screening. That’s um … when she’s pregnant and they take the tests, so that’s when you’re tested for all the other diseases, it will show up then if the child, is going to have sickle cell. […] Well I don’t know, because from my understanding, it’s um … you’re taking this test in newborn stages … I mean in ah … prenatal stages, right. So obviously, while you’re pregnant, your doctor is mandatory, just like Tay Sachs disease, is going to be doing the blood work on you, and on the blood work it’s going to show up what you’re potentially carrying, having, whatever whatever, right? So that’s my understanding of it, maybe not, but I mean that’s what I was told that while you’re pregnant you’re being tested, right, and this will screen and it will also tell you all the potential. (#28, advocate)
SO, the issue isn’t what’s the balance of interests today, the issue is actually what’s going to be the balance of interests between the best, the very best interests of the neonate, the unborn neonate uh … versus the level of knowledge and understanding of a parent to give truly informed consent. I wouldn’t have a problem if the incent…if the consent or dissent is truly informed. I defy you to show me a case study of a parent who has dissented who has a full understanding of the risks they are assuming for their unborn child. (#2, advocate)45

While future research would need to be conducted to ascertain the prevalence of such confusion within the province between prenatal and newborn screening, that three of my participants deemed key informants for NBS in Ontario, having participated and contributed in some way to the expansion of the program, confused prenatal and newborn screening suggests that the possibility exists for parents less connected and less informed about NBS to confuse the two.46 All that to say, as Ontario considers how to proceed with consent for NBS the possibility for confusion and/or misunderstanding between prenatal and newborn screening is real and should at least be taken into account moving forward.

Ultimately, the fear that lies at the core of these logistical concerns pertaining to explicit informed consent is that a mistake will be made inadvertently and an infant who would otherwise have been screened is missed and ends up being affected with one of the conditions on the panel. A few participants articulated that errors may be introduced at the laboratory level as a result of an added layer of decision-making introduced by an express consent option. The quotation below provides an example of some participants’ perceptions of potential errors that can be introduced if consent is sought for storage and research:

45 This participant is very knowledgeable on NBS and knows the difference between NBS and prenatal screening. However, I have included this excerpt simply to underscore that the possibility for people to make a slip and confuse the two, even among those considered extremely knowledgeable on the subject, exists.

46 To underscore the possibility further, a study by Davis et al. (2006) also found that some of their participants “confused newborn screening with testing for jaundice, prenatal laboratory testing, and hepatitis B immunizations” (p. S330).
And then when people are receiving the specimens they’re looking at the boxes and then they will decide is this for discard or is it for long-term storage. So then, you have an extra layer here at the decision making at the time of the specimen receiving. [...] So when the specimens are received in the lab. So they have to sort two piles, the discard and storage for long period. So, this is decision-making (participant raps on the table) from people that have not been trained to do so. [...] If the technicians have not been trained to do so (participant raps on the table), they will continue like they’ve been doing forever, and forever here in Ontario is keep for long. So they don’t have to be extra careful at knowing where the specimens have to go after. [...] Because it’s one thing at the time of specimen collection (participant raps on the table) to do the consent, but then it’s the handling of the specimens. (#17, advisory committee)

Errors may also occur amidst the hospital chaos if a parent accidentally forgets to sign a form or HCPs forget to present parents with the form:

Well, I think that even if just a positive consent form had to be signed for every single family and it had to be somehow tracked. Because if you have a form that's one thing but there has to be some kind of process to make sure it's actually done. I suspect that might result in, you know, just imagine a busy labour and delivery ward, they're discharging 10 babies one day and the nurse says, oh we don't have the consent yet for this baby and so we can't do the screening and then things get busy and somehow the baby gets discharged without the screening being done. (#10, advisory committee)

“It [express consent] could ... it could be done at the same time as the education process but, then again, there could be barriers to the whole informed consent process because, what if you get a situation where a parent just mistakenly forgets to sign a form […] (#46, advocate)

Regardless of how the oversight occurs, the unintended outcome is the same: a baby who would have otherwise been screened is missed. Even within the current practice of implied consent for NBS mistakes occur:

I specifically had asked her [the nurse]. I said, ‘Oh, are you going to do her newborn screening test at the same time?’ And she said, ‘Oh, no. Not now.’ I said, ‘Okay, fine’ and then, when she came back with my daughter, she handed me this slip of paper [the requisition form] that had the number for the newborn screening test and she said: ‘Oh, here you go. This is for her newborn screening test.’

I said, ‘Oh, I thought you said you weren’t going to do that right now’ and she said, ‘Yes. Well, I changed my mind.’ Then, of course, I noticed that my newborn didn’t have a
Band-Aid on her foot and I said, ‘You did the test but you didn’t take the blood from her foot?’

‘Oh, no’ she said. ‘I took it from her wrist while I was doing the other blood draw.’ Oh, really. OK. Hmmm. So that kind of surprised me. So, ironically, just a short time later I actually had a call from a metabolic specialist and he was asking me how things were going and stuff and I happened to mention to him that they did the newborn screening test, but I was surprised they took it from the wrist; they didn’t take it from the heel. He didn’t say anything and we got off the phone and within five minutes the charge nurse came in and she said, ‘[Participant name], you must have misunderstood the nurse.’ She said, ‘the heel prick test, the newborn screen test wasn’t done on your daughter.’ She said, ‘You know, I looked at the chart and it doesn’t say that it was done.’ And I said, ‘Really?’ I said, ‘Well, if it wasn’t done, why do I have this?’ (referring to the NBS requisition form) and she looked and she was quite upset because A it hadn’t been charted and B it wasn’t done correctly. So she actually took the baby and redid it. So I actually have two pieces of paper with different serial numbers on it for my daughter.

But you see, if I was not fully informed, this is how mistakes can happen because A that particular nurse, obviously, doesn’t understand the importance of a newborn screening test and B is not fully informed as to what the test was all about. Because, if she was, she would have taken the situation much more seriously. (#46, advocate)

As participants discussed express consent for NBS in Ontario it was apparent that they had reservations based on their perception of what would have to be conveyed, who would be responsible for conveying it, and how it would be operationalized. I interpreted these concerns introduced through an express consent process as a potential harm to the goals of the screening program, namely NBS uptake. However, many participants also voiced their concern that express consent would not achieve its conceptual goals of enabling informed, autonomous decision-making. I explore this theme in the next section.

*Analytic/Interpretive Theme 2. The Challenge to Autonomy: Harms through the Ritualization of Express Consent*

I have framed my participants’ perceptions of and attitudes towards express consent for NBS as a tension between achieving population health goals and protecting individual
rights and freedoms. The first overarching analytic and interpretive theme focused on presenting and interpreting data that focused on perceptions of express consent as a potential harm to the goals of the program. This major theme, “The Challenge to Autonomy: Harms Through the Ritualization of Express Consent”, examines participants’ perceptions of challenges to the effectiveness of express consent as a least restrictive alternative to NBS in terms of its ability to foster informed, autonomous decisions. As I articulated throughout Analytic/Interpretive Theme 1, there is necessarily some overlap between dimensions of an express consent process that are perceived to threaten NBS uptake that can also be interpreted to threaten parental autonomy. For instance, if information is presented in such a way that parents are overwhelmed by and/or misunderstand the information presented to them and then they decline screening, one can reasonably conclude that they have not exercised their autonomy. Similarly, the role of HCPs can either support or undermine parental autonomy, depending on how knowledgeable the HCP is and how the information is presented. In addition, if express consent is implemented in a way that privileges some members of a society but not others, then the rights and freedoms of some parents, as conveyed through autonomous decision-making, are arguably compromised as well. Finally, pragmatic and logistical barriers to implementing express consent can also simultaneously affect NBS uptake and parental autonomy, since unintentional oversights and mistakes can undermine parental decisions.

Building on this notion that express consent could compromise parental autonomy, another theme I identified and interpreted in the data on express consent pertained to some participants’ perceptions that the theoretical goals and ideals embodied in the concept of
consent (e.g. informed, voluntary, non-coercive, autonomous decision-making) may not be realized in practice. I identified five sub-themes that work together to support an interpretation of some participants’ perceptions of express consent as not “real,” not “true,” not “legitimate”—a mere illusion of choice rather than a reflection of informed, autonomous decision-making: a ritual. The five sub-themes include, 1) “True” express consent requires knowledge and understanding; 2) The timing of express consent can impede knowledge and understanding and/or be coercive; 3) “True” express consent cannot be coerced 4) Resources are essential to achieve effective express consent; and 5) Doubts that parents really give “true” express consent in jurisdictions that require express parental consent for newborn screening. I have interpreted these findings collectively as a perceived ritualization of express consent: a harm to the integrity of consent as a concept and ultimately a challenge to achieving autonomous decision-making in practice.

“True” express consent requires knowledge and understanding

Some participants identified knowledge and understanding as integral to any express consent process and critical in helping parents make truly informed, autonomous decisions; yet they also questioned the ability of express consent to achieve this goal in practice. Specifically, a number of participants challenged the extent to which parents could ever fully understand the complexity of NBS and the 28 conditions on the panel. Participants further questioned whether parental consent given for NBS without full knowledge and understanding could be considered “true”, “legitimate”, or “real” express consent.

The information that many participants felt would need to be disclosed within the context of an express consent process was considered complex, confusing, and rife with
possibility for misunderstanding. Not surprisingly, when considering the achievability of “true” express consent, participants immediately honed in on the information component of consent and stressed that, “true informed consent” requires understanding:

You know, there comes a point where you’re saying, ‘is this real consent or not...or is this not?’ If you’re consenting to a particular thing that may have significant implications, particularly say a research study that is very different from the normal therapy, you know obviously you need consent but still it’s still always a question of, how ... cuz consent requires informed...it’s informed consent, it’s not just consent, it’s informed consent. Well what’s your understanding of the information that’s been shared with you? (#50, treatment centre)

But some of it is, you know, that's the challenge, right? It's to make it [NBS education] so that it's very easily accessible to people. And the same thing in terms of informed consent. If you don't put the information out to people in a way that they can really understand it you're not really getting informed people, you're getting people that are just either frightened by it or coerced into it or they just don't understand. (#41, advocate)

I do support that principle, that it’s better to do it [NBS] with an informed choice than making things legislated or mandatory. So in principle I agree with that. My conflict with it is, if you’re not truly giving an informed choice what are you doing? Do you know what I mean? So if we’re … with Sickle if there … if there were going to be um … an info … people having the choice whether they want to find out about their child’s carrier status, let’s say, I mean that requires a conversation; so if you’re not really having that then is it informed choice, you know what I’m getting at? It has to be either elements that are required if it’s going to be true choice, and ah … either I think we either have to enable that to happen, provide the materials that allow that to happen so both educational and, you know, provider education and time so that that actually happens. Or, I’m questioning whether we’re really doing it. (#3, advisory committee)

These quotations encapsulate many of my participants’ skepticism surrounding express consent in terms of its ability to achieve its theoretical goals in practice, particularly as it pertains to ensuring parental understanding. Participants underscored their beliefs that achieving informed parental decisions requires providing the information in a way that supports and facilitates understanding—creating the conditions necessary to foster and
promote self-determination. Among the conditions perceived by my participants as necessary to establish parental knowledge and understanding for NBS include providing appropriate educational materials not only for parents, but also HCPs; allowing sufficient time to educate parents; and to present the information in a way that will support informed decision-making. The timing of the express consent processes was given particular attention by my participants and, therefore, is discussed in the sub-theme below.

The timing of express consent can impede knowledge and understanding and/or be coercive

The question of when to seek express consent is a challenge familiar to NBS programs and a dilemma frequently discussed in the academic literature. Unsurprisingly, the challenge of timing was also on the minds of the participants interviewed in my study, particularly in terms of its ability to influence parental knowledge and understanding. In particular, many of my participants expressed concern that seeking express consent for NBS post-labour and delivery – otherwise “stressful” situations – would impede parental knowledge and understanding, thereby rendering it an inopportune time to obtain “true,” “legitimate,” or “real” consent:

You know, can you have an informed consent from 150,000 women who are in a fairly stressful time of their life where they have an obstetrician, you know, you're getting delivered… you're delivering your first baby or your second baby. Like I mean, you could argue is it eve… is it informed having somebody sign that, you know the 24 hour 48 hour period after you've delivered? And can you really expect people to understand screening for 27 different disorders beyond a… a certain fairly basic level, okay? (#15, advisory committee)

So, this is why I said a kind of consenting mechanism that gives the family the right to say, (participant bangs on the table), ‘We really … we do not want to have this done,’ this is what we definitely need. But, a kind of informed consent system that is not working in reality that makes no sense. The point is if really parents should give an informed
consent they would need proper information. So … and when should they get this and who would give the proper information. It’s impossible to give this information one day after birth, so that’s a time when they have other things and they couldn’t really listen to someone who tells them about 30, 50 diseases. So, it would have to be done during pregnancy, and there is no system in place. We need to come to provide them with the information that they would need. (#36, treatment centre)

And the issue, too, is that right after birth, families are often quite exhausted and tired and I guess it's often difficult for them to take a lot of new stuff in, too. I hate to say that, but it is true. Women are often a bit anaemic, they're tired, there are a lot of physiologic changes happening at the same time. They're just exhausted (participant laughs). It's not the greatest time to really try to absorb a really complex new thing. (#31, advisory committee)

A few participants described their perceptions of how little thought is often put into express consent processes:

[…] I think they’re [women47 are] incredibly vulnerable at that point: you cannot achieve informed consent under those circumst…truly informed consent until they’ve had time to reflect on it and not just, ‘oh, I’ve given a ten second thought, fine.’ You know, (participant laughs), that’s not informed consent. Not when it could mean the difference between life or permanent, severe disability. (#2, advocate)

Combining what participants perceived to be a substantial amount of complex and potentially confusing information about NBS in a cumbersome express consent document and administered at a stressful time such as post-labour and delivery was considered a prelude to an uninformed decision. For example, if new mothers and/or their partners are presented with what may be perceived as a labour-intensive express consent process, they may either “consent” without fully understanding what they consented to or refuse screening merely to avoid what they may perceive as a cumbersome, burdensome consent process. In short, one might interpret the timing of express consent as potentially coercive in its ability to either facilitate uninformed consent or uninformed dissent. Participants also identified their inner-

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47 Participant’s qualification.
conflict around the possibility for coercion in express consent in terms of the way in which information is provided to parents: simultaneously recognizing that express consent cannot be coerced, yet wanting information presented in a way that would induce parents to consent. This theme is explored below.

“True” express consent cannot be coerced

Participants were clear that for express consent to be considered “true,” “legitimate,” and “real” parents could not be coerced into consenting to NBS. However, as explicated earlier in this chapter, there was also the perception among some participants that parental consent could not interfere with the larger NBS program goals and objectives: namely to ensure that all babies are screened. Therefore, with this in mind, some participants felt that the information connected to express consent for NBS could not be neutral, but rather needed to be presented in a way that would encourage parents to consent:

So, (pause) of course they need to know, okay, what will be done with my child. So, the heel prick for example, they need to know this. But, I mean, the information should have the intention to convince the parents to say yes, you know, that’s the point. So it’s not a (participant’s rapping on the table) neutral information. Because we had a participation rate of newborn screening over the last four decades, if the system was working well, that was in the range higher than 99%. So, if we get lower agreement rates because of extended information then I would say there is something wrong, so then the information that we give is not appropriate. So, to make it more successful, we would have to show them how serious the consequences would be, but on the other hand this is what nobody wants to do. I don’t want to scare them about diseases the child is having with a chance of one in 100,000. But, in fact, that’s why we are doing the screening, you know. (#36, treatment centre)

While scare tactics were not recommended, a few participants acknowledged that discussing the implications of not screening are considered scary. However, if parents are motivated to
consent out of fear, participants questioned the extent to which such consent is “true” informed consent:

[S]o if you put something like that [a decision aid to facilitate informed consent for NBS] in place, would people stop having newborn screening (participant asks rhetorically and laughs)? You know, like what would happen (participant still laughing) … it might not … not accomplish what we really want which is informed decision making but truly informed … like truly understanding what it is … Like if you just scared them that wouldn’t be the point, you know what I mean? (#3, advisory committee)

Within this apparent tension between parental autonomy and screening uptake, some participants felt that if express consent would ultimately be designed in a way to induce parents to accept screening and discourage parents from declining screening on behalf of their infants, then why frame it as a choice or even introduce it at all:

You see, if your final, final intuit is that it is not okay for anybody to say ‘no’ (participant bangs on the table repeatedly), then you can't have applied, you know, just a … you know, you've got to go through an absolute process. And I think that to me is kind of what this is all about. So if you would ever say, ‘yeah, sure,’ a family can kind of go through all the risks and benefits and they will feel it's okay for them to reject it [NBS] on the basis of whatever, I would say sure, you could [unintelligible 42:41/sounds like “take it [informed consent]”]. But I would say that ‘no,’ at the end of the day, you don't really want to give that option. You don't want to leave it open that a family could turn it [NBS] down, except if there's some really deep-seated religious belief in there. And even then, one would have to seriously challenge it, right, when you look at some of the other kind of religious practices that could put children at risk. We don't allow those either. In this one I would say it's debatable as to whether or not you might want to do it […] So why give a person a choice if you don't really want to have a choice? (#41, advocate)

This particular sub-theme in many ways epitomizes much of my participants’ ambivalence towards express consent for NBS. Participants appreciate the importance of an express consent process for parents that is free of coercion, but worry that the very presentation of the potential implications of parents not pursuing screening for their infant – however rare – is in itself coercive. And, to the extent to which NBS information could be presented in an unbiased way, participants questioned whether such an approach would be
good for infant – and public – health. Ultimately, participants questioned whether an approach to express consent for NBS that encourages screening uptake but does not fully support parental autonomy should be implemented. These tensions contribute to one of the conclusions of this thesis, namely that future approaches to consent for a medical public health intervention such as NBS will have to strike a balance between attaining the goals and objectives of the public health program and fostering self determination.

**Resources are essential to achieve effective express consent**

Participants also identified appropriate and sufficient resources as an integral component towards ensuring the implementation of a proper express consent process that achieves its goal of generating informed decisions. However, at the time of the program’s expansion Ontario purportedly did not have such resources:

> [T]he consent issue becomes a **bigger** issue because we … we realize that consent for screening, uh the testing in the **first place**, had uh major resource implications. It takes **minimum** 20 minutes **per person** to obtain **decent**, not even **decent**, but any kind of consent and uh on the basis of the number of… of people **involved**, that is clients or **patients**, uh the **nursing time** involved, assuming the nurses would be obtaining the consent, was **prohibitive** uh… to… considering the resources we [inaudible 18:44/sounds like: ‘have in hand’]. So the decision to say **insist on**… on an informed, written, signed consent to screening ah in the **first instance** had major resource implications which we uh were not uh equipped to address. (#54, advisory committee)

Implementing express consent without such resources was perceived by some participants as having potentially negative implications for the concept of consent, parental autonomy, and screening uptake, thereby threatening to make a “mockery” of the whole process:

> Um (pause), I think you know, if we really had a lot of **resources**, it would be wonderful to sit down and explain everything. But, unfortunately we don’t have. So then it just, kind of, becomes a mockery, as I told you, you know. I don’t think that mom [who refused NBS on behalf of her child who was ultimately affected with one of the rare conditions on the screening panel] understood, what it [NBS] was. So, then the best
option would be like what we have in terms of, you know, implied consent. (Pause). That, it’s not ideal, but I think that’s the best, because you cover, at least, everyone. (#53, treatment centre)

But I think, again, in an ideal world (participant raps on the table) you would have the resources for decent education provided in a way that it’s understood, truly informed consent, explicit, opt in, opt out, sounds great. It’s very utopian. (#42, advisory committee)

One participant described a similar concern regarding the discussions around how to disclose carrier status for sickle cell anaemia. Purportedly, among the issues discussed included questions of resources, feasibility, and whether “authentic choice” can occur in practice.

This participant questioned the extent to which “optionality” should be pursued if it cannot be implemented “properly” or succeed in “accomplish[ing]” its objectives, namely “authentic choice”:

So we sort of created some options um…and criteria were um…the alignment with evidence, alignment with other jurisdictions,…cost, liability…(long pause) those were some of the key criteria we used to try to land on the most reasonable solution. Um…but it wasn't easy because if maximizing choice was important, feasibility would make that impossible. So if…if…if we insisted upon a full consent model to getting these results, the…the providers on the ground that the system would depend on to accomplish that, would for sure resist the imposition of that direct policy direction to the point that it probably wouldn't be feasible. So it was a balance between what in principle should happen. Again, and what realistically can happen. And, you know, then the sort of question of ‘Well, if we can't get … do choice and "optionality" properly, should we do away with it altogether?’ You know, if we…if we can't accomplish what we think is…is is authentic choice…, then what's the point of even trying? Maybe we should just mandate this and take "optionality" off the table. So that went through a…, you know, everything went through its sort of round of discussion um consideration. (#27, advisory committee)

This sub-theme underscores the perception that an appropriate infrastructure is necessary to support an effective approach to express consent for the various dimensions of NBS, from the heel prick, to carrier status disclosure, to storage and research. While
arguments may differ as to whether such consent is necessary for the various dimensions of NBS, the perceived challenges of ensuring that express consent achieves its goals of honouring parental autonomy and obtaining true, informed, autonomous decisions are similar. Participants argued that an ineffective consent process for NBS, resulting from reasons including insufficient resources, would make a “mockery” of the theory of consent in general. I have interpreted such concerns within the broader thematic framing of consent as harm-causing with particular implications for autonomy (although a financial expenditure for a sub-standard process that may not achieve its goals as a least restrictive alternative arguably have implications for population health as well). If express consent as a least restrictive alternative for a public health intervention is ineffective, one might ask, who benefits from a ritualized approach to express consent that merely offers an illusion of choice? One might also ask, particularly given the public health nature of the program, what are the risks of perpetuating such an approach and, perhaps more importantly, who bears those risks?

**Doubts that parents really give “true” informed consent in jurisdictions that require express parental consent for newborn screening**

The final theme to support my interpretive framing of participant perceptions of express consent as a ritualized practice rather than an earnest effort at achieving “true” informed consent was some participants’ skepticism regarding the kind of consent obtained in jurisdictions that currently require express parental consent. Some participants’ reactions to these jurisdictions’ processes and high uptake rates appeared to be reactions of suspicion. Specifically, participants questioned whether these jurisdictions simply go through the motions of express consent rather than achieve true parent understanding followed by
informed decisions. Put simply, are informed, autonomous decisions being made while preserving screening uptake, or is express informed consent in these contexts really more of a ritual geared towards screening uptake?

For instance, a few participants referenced Maryland, Massachusetts, and Wyoming, three US states that required express consent for NBS at the time of my interviews. Some participants wondered whether these programs truly encourage careful, considered reflection or is the process of “informed consent” really more “routine”?

Another point is that while it's [NBS’s] described as optional and voluntary [in Massachusetts], how does it really look? Like, are people really making careful choices? Or is it kind of being rolled into something routine? You know, are people really being engaged in a conversation about what this means? Or is it kind of volunt…you have to know to opt out and that's how it's voluntary. I don't actually know. I think in Massachusetts it's um…I think it's a tick box and I think…I don't know the conversation that precedes the tick box if it’s a very detailed conversation about the risks you’re getting yourself into by engaging in newborn screening or if it’s a much more mechanical um…tell us if you don't want it. But then…people need to know why they wouldn’t want it and so they don't not want it because they don't…there is no good reason…explained to them why they wouldn't. I don't know how it actually works. (#27, advisory committee)

Another participant felt that these jurisdictions must present the program “really well”:

Well I know that that’s [consent’s] ethically the way it [NBS] should be done and I know that in Maryland and Wyoming that their refusal is pretty low. Surprisingly low, actually. I was amazed at how low it was. It may have been presented very well in those states […] (#5, advisory committee)

One participant described another jurisdiction that had structured their NBS program around a model of written, express consent, yet screening was purportedly still performed on all samples without checking to see whether consent had been obtained. Behind the veil of express consent, the program was perceived to operate according to an implicit assumption that parents have in fact consented:
[This jurisdiction now has] informed consent, written and informed consent. The thing (participant laughs) ... the trick at the end leads to a participation rate or to a screening rate that is still much higher is nobody really checks whether the consent is there (participant says slowly and deliberately). You know what I mean? There needs to be a consent and the consent is ... okay, so it’s not true for all programs in [the jurisdiction] because [they] have a similar system like Canada. But, in general, there is a requirement that the family gets the information about newborn screening and signs this before the test is done and the signature goes to the child of the mother. So, mother’s patient chart. So, if someone would question later you did this without our consent, then they could show. But this consent does not go to the screening laboratory. So, the screening laboratory could only, in principle, only do tests if they have the written consent. But, this would delay the newborn screening, you know. So ... And this is why most of the programs, at least two years ago, I’m not quite sure whether [they] have changed this now, nobody was checking whether there was a consent and the test has been done. And it was the understanding that the signature is in the chart. And then there was a survey checking whether it was true or not and it was true in 80%, but it was not true in 20% (participant says with a smile). If you strengthen the requirements and you would say, you can only ... the laboratory is only allowed to do the test if there is a written consent, this would cause, I guess, a decrease of the completeness by at least 5% to 10% and this would put children at risk not to be detected with a disease. So, this is not a primary goal but this is one of the prerequisites of newborn screening is completeness. (#36, treatment centre)

Although jurisdictions that require express parental consent for NBS report high rates of NBS uptake, some participants remained concerned that the potential for declining screening is increased through an express consent process. And however theoretical a risk of refusal might be, that was not a risk Ontario wanted to take:

There is certainly evidence from Massachusetts that is...much of it is option based and pilot based. And...I think they have 99% uptake. So...one issu...one point is that it's a theoretical risk that people aren't willing to contend with. Even if it's theoretical and even if in Massachusetts, in the U.K., its been demonstrated to not be a problem, no one here could take that risk. (#27, advisory committee)

In addition, while other jurisdictions may have found – or at least created the perception of – a balance between parental autonomy and screening uptake, a few participants articulated

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48 My interview with this participant took place in April 2010.
their belief that success in one jurisdiction could not necessarily be transferred to Ontario given perceived cultural differences:

I think that there is some research necessary in this area [of express consent for NBS] and some of the research might simply be reviewing the literature and reviewing what other jurisdictions do. We haven't done that yet. Uh, one has to take into consideration cultural differences uh and particularly when you try to extract … apply UK uh methodologies here we are working in a different cont…social context. (#54, advisory committee)

Drawing on participants’ perspectives of and attitudes towards express consent, if “true,” “real,” and “legitimate” informed consent cannot actually be obtained in practice through an express consent policy, then the concept of express consent is reduced to merely an illusion of choice that may neither achieve its goals of fostering autonomous decision-making nor its public health objectives of ensuring high screening uptake. Interpreting express consent as a ritualized practice that may actually cause harm to parental autonomy challenges the policy and practice of express consent as an effective least restrictive alternative in terms of its ability to elicit informed, autonomous decision-making. Drawing on the public health ethics lens further, I have interpreted these analytic/interpretive thematic findings as a challenge to express consent’s ability to achieve the balance between both respecting individual liberties and achieving the goals and objectives of Ontario’s NBS program. Such perceived shortcomings of the practice of express consent for NBS – as identified by my participants – ultimately raises questions worthy of future exploration about the potential effectiveness of express consent as a least restrictive alternative for NBS in Ontario.
Analytic/Interpretive Theme 3. A Search for Balance: Reconceptualizing Express Consent Within the Context of Newborn Screening

In this chapter I identify and interpret perceived tensions related to express consent for NBS and ultimate ambivalence among many participants who appear torn between wanting parents to be better educated about NBS and to have an opportunity to make informed healthcare decisions on behalf of their infants, yet also fearful about what such parental freedom could do to the provincial NBS program’s ability to achieve its public health goals. In this third analytic/interpretive theme, “A Search for Balance: Reconceptualizing Express Consent Within the Context of Newborn Screening”, I draw on data from my transcripts wherein participants spontaneously discussed their perceptions of and ideas for different types of approaches to consent for NBS. Contextualized within the broader finding that many participants are skeptical of express consent’s ability to realize its theoretical goals in practice, I interpreted my participants’ efforts to identify alternative approaches to consent for NBS (or additional practices that could better support and facilitate informed decision-making) as an attempt to identify (or an interest in identifying) an effective approach to NBS that would work towards bridging the gap between individual rights and freedoms on the one hand and the broader societal good on the other. In the pages that follow I demonstrate that although some participants were adamant that express consent for NBS is not necessary given its public health nature, others were equally adamant that express consent is necessary, and many others appreciated the tensions between the two and conveyed ambivalence. Considered within what was perceived to be a changing landscape both in terms of NBS and parent expectations, I provided my participants’ thoughts on three possible approaches to consent for NBS: incorporating a check box component to express
consent; a simple consent approach; and/or a decision aid to support parents through the informed decision-making process.

**Perceptions of the need for express consent for newborn screening**

For some participants, the very need for an express consent process within the context of a public health program was considered “overvalued”:

> I think that people want to make sure that everything’s done correctly and that’s laudable, but I also think that at a certain point you become overly concerned about potential objections and it stands in the way of doing *more good for more people*. The question of choice and option is something that is offered at every opportunity now, but it comes with some cost too in a larger public health sense. [...] but I think consent is often — I mean, it needs to be looked at, it needs to be done properly, I don’t have an issue with that. But from the standpoint of *population screening* where you have the public health perspective, I think that the importance of that process is *overvalued* compared to the benefit of the population as a whole. (#11, advisory committee)

Others, however, felt that “it’s always better for people to go into testing with their eyes wide open” (#12, advisory committee) and that ultimately seeking consent is “*ethically the way it should be done*” (#5, advisory committee). One participant even stated that express consent probably should have been instated when the program was first implemented in the 1960s:

> [Y]ou see *consent and the revised program of the newborn screening are two different things*, because consent should have been *in place* since they started the newborn screening in 1960-something. So, consent has been something that has been *weakly done* for years and years and years. (#17, advisory committee)

Such arguments endorsing express consent for NBS were bolstered further by some participants’ perceptions that ultimately patients (and parents in the case of minors) expect to be consulted with, educated about, and given an opportunity to consent to the health care procedures or interventions that either they or their children will receive:
Yeah, because *we don't really have a consent*. You know I think that was *fine* until more recently. But I think more recently society has changed, I think expectations have changed around consent and *patients' expectations* have changed around how medical procedures are explained to them. And I think the public's expectation is much *different*. And so *I think it* would be better to move to some kind of a more formal consent process. (#10, advisory committee)

*Certainly especially* if it’s going to expand *anymore*, and start producing more *common* results; you know, if people are going to start getting more and more results, we need to educate both sides much better about what it is … cuz I think that’s what people *expect* in medicine. (#3, advisory committee)

Rendering NBS in Ontario mandatory appeared to remain, at least for some participants, among the possible options for Ontario’s NBS program moving forward; a perspective seemingly rooted in the philosophy that NBS is ultimately for the broader public good:

I think there was a *lot of discussion*. I think there was considerable discussion of how one would *mandate* newborn screening. I mean, let’s remember that the whole *process* is for *the public good*, and so I would say that a lot of this discussion was very appropriate, very altruistic, one wanted to do the best job *possible*. And I think that there has been *continuing dialogue* about this that has expressed *concern* that if we make the process any more complicated than it already is that there’s a real *fear* that families would *opt out* of the program. The concern being that that would put kids *at risk* of not being *ascertained* and therefore not being treated. And so, I think that *most people* believe that as a *public good* one should make this as easy as possible. (#42, advisory committee)

Although there was a sense that mandatory screening remained on the table, a few participants acknowledged that it would be preferable if the public health goals could be achieved through voluntary measures:

*Flipping* the … the *opposite perspective* would be to say, let’s make newborn screening *mandatory*: it’s a *mandatory* part of newborn care, and you *don’t* have the right to dissent and you don’t have to give *consent*. And ah … ah … that’s … that’s … I think that’s *again*, it’s *not the* … I guess if there’s a general *philosophy* I think they tend to *not want* to make things *mandatory* (participant says with a smile), but I think that’s [mandatory screening’s] an option. (#49, advisory committee)
Although voluntary approaches were described as preferable, at least in theory, participant perspectives and attitudes towards implied and express consent reflected skepticism that either approach could successfully achieve the public health ethics balance between ensuring that public health goals are attained while also protecting individual rights and freedoms. I identified the unspoken question as, how can this balance be achieved?

Some participants who discussed the possibility of express consent were adamant that such consent would require a full detailed explanation of all 28 conditions, plus storage, plus research. However, such an undertaking was perceived not only as impractical and untenable at a systems level, but also overwhelming and confusing at an individual, parental level:

**Respondent:** It [express consent] means where someone actually sits with you, goes through with you in great detail everything and then you have to sign some sort of consent form saying yeah, I understand this and this is what I want done. And oh, I want this test done but not this one and I want that test done but not this one. So I want CF but I don't want to know about sickle-cell and I don't need whatever. Sure, I'll have PKU but not thyroid—

**Interviewer:** Okay. So there's a sense in your mind that an explicit consent would be, parents would know all 28 conditions.

**Respondent:** It would have … Yeah. I mean I think that if you're going to go to an explicit, like a signed consent model (participant raps on the table), then that's what you have to do. You have to have someone actually sit and explain it all to them—what it means, what is the outcome, what are the implications. I just think that would paralyze our system. And confuse people, too […]. (#31, advisory committee)

Many participants spoke against “a detailed laundry list of choices”, arguing that such an approach would be neither “feasible” nor “reasonable” (#27, advisory committee).

Moreover, participants were concerned that introducing express consent through a detailed, comprehensive informed consent process could confuse parents and create misunderstandings and possibly lead them to decline screening on behalf of their infant
altogether, in so doing putting infant health at risk and threatening the goals and objectives of
the program — a fear many participants did not want to see actualized:

It seems to me that sometimes people see newborn screening programs or other sorts of
public health programs as being so important and so … um … helpful and beneficial that
any potential alarmist concerns that are raised might threaten the existence or the success
of the program.⁴⁹ So I think … um … if … if … ah very remote risks are explained to
families they … the remoteness of the risk might not be appreciated and so consent
might be refused on that basis, and so the program itself might be in jeopardy. (#1,
advisory committee)

As participants talked about express consent possibilities for NBS in Ontario and their
concerns that express consent may neither work well for parental autonomy nor program
uptake, a few participants discussed three possible alternative approaches to consent for NBS
that they perceived could potentially better include parents without compromising the public
health goals of NBS in Ontario: a check box approach; a “simple” consent; and decision aids.
Given my public health ethics lens of analysis, I interpreted perceived possible alternatives as
efforts to reconceptualize express consent within a public health context in order to achieve
the balance between uptake and choice.

**The check box approach to consent**

A number of participants introduced the topic of a check box as an approach to NBS
that some jurisdictions have adopted whereby disorders are broken down individually or
according to condition groups and parents can then consent to each condition individually or
each condition group:

⁴⁹ It is important to underscore that program success is defined among many of my participants as identifying
and treating every baby affected by one of these conditions (#15, advisory committee).
I saw a poster at a scientific conference about … they have an informed consent/dissent, so called “policy,” in Scotland for newborn screening. As a matter of fact, the form is so specific that you can say yes to congenital hypothyroidism and no to PKU. It’s … it’s condition specific, all right. (#2, advocate)

I’ve seen or heard some places talk about breaking it down into these are conditions where we can prevent mental retardation, these are the conditions where we might prevent ah sudden infant death, that type of thing that might be an easier way to do it [inform parents]. (#20, treatment centre)

However, a number of participants expressed skepticism that such an approach to express consent for NBS succeeds in escaping the concerns articulated earlier in this chapter (see Analytic/Interpretive Themes 1 and 2). Specifically, does a “check” reflect parental knowledge and understanding, or is such an approach to consent merely different packaging creating the same ritualized, illusion of choice:

I mean (participant raps on the table), when we had debated this, these same topics (participant raps twice on the table) before, you know, the question of should we have a check box list on the screening card, you know, do I want this, do I want this, right? I mean that was an idea: do I want secondary storage, do I want carrier disclosure. In the context of a woman giving birth and all the other stuff going on, you know, I think the probability that you’ll get an informed decision or an informed choice is pretty low. The check box most likely […] there is a question of] whether or not it [the “check”] actually does reflect what their choice (participant raps on the table) would have been if they actually understood, you know. (#18, advisory committee)

I think the tick…the value of the tick box is that it’s an enforceable, easy mechanism (pause) and looks like choice. The concern is that it may not mean choice. It’s very hard to know what goes on before the box is ticked or not ticked. And I think this province isn't comfortable with endorsing an approach that has a black box behind it. It has a tick box, but what goes on in order to establish that tick is not clear. And I think there is a major fear of it…of any process that would be in place to support a tick box could also jeopardize the uptake of the program. (#27, advisory committee)

[A] description of a Dutch program where there was an opt out box for whether or not families wanted to receive incidental results like sickle cell carrier status. And at the end of a qualitative research program, they [the researchers] got a sense that parents didn’t understand (participant laughs) the opt-out properly. (#42, advisory committee)
In addition to questions about whether express consent would be “truly” achieved using a check box approach, one participant introduced potential logistical challenges of such a consent method. Specifically, while conditions might be easy to separate on paper, a couple of participants questioned whether such separations are feasible from a technological, laboratory standpoint. In addition, the check box approach raised additional ethical dilemmas regarding the incidental findings an ongoing challenge for NBS in Ontario:

One option … so that's the… the flow of the discussion o… on consent and then the… the issue was, you know, other questions that come up. Well sh… you know, should we have a tick… a tick-box type of approach? You can screen for A, but not B. C, not D. I consent to research or not. I consent to storage or not. I consent for data collection or not. Again, one of the… the I guess the principles is that we don't want to introduce something that would negatively impact on the program. And… in this instance what I mean is, in… in my view, negatively impact on the benefit to the neonates in the province. I don't mean the program in the sense of some system entity. I mean in terms of the success in identifying kids who needed to be treated. And my sense, and this is an opinion, was really that if we started having a lot of options, then you would erode into … maybe the uptake of the testing. And I also, at what stage, it's… do you stop asking for the tick-box, okay? I mean there are other practical pieces about that. I mean being able to test for A or B may not be practical at the lab level. I mean, some of it is, but some of it’s not, okay? And does the person when they tick A and not B, do they really know that we've got that data. But they are now [inaudible 1:05:30/sounds like: ‘up and’] saying they don't want it ever, okay? So there were issues around that. (#15, advisory committee)

Finally, in keeping with the overarching theme of this chapter – the perception that express consent could cause harm to autonomy and infant/public health – an unanticipated “finding” emerged through the process of consent for my study. As mentioned in Chapter 4, I incorporated two check boxes in my consent form followed by a signature (see Appendix F). These check boxes proved extremely challenging throughout my recruitment. Participants often skipped over the check boxes entirely and went straight to the signature, returning to the check boxes only after I drew them to their attention. Despite my vigilance,
two out of 57 consent forms were signed but the check boxes had been left blank. The conclusions that can be drawn from this unexpected occurrence are extremely limited, as this was not a study about the number of consent forms that slip by without check boxes filled in. However, in a thesis that underscores participants’ perceptions of express consent as potentially harm-causing with a particular focus on logistical obstacles and the potential for error and the perceived magnitude of such error, this small example in my study does underscore the reality that the possibility exists for unintentional oversights. As one participant stated, in relation to his/her recollection of the Dutch NBS program who had implemented a check box consent for incidental findings, even when you think you have implemented a consent approach perfectly, it is never as perfect as you would like to think:

> And so, even in circumstances where they [the Dutch NBS program] felt the program was being delivered well [via a check box approach for incidental results], as usual, it isn’t done as well as you think and you don’t get the same kind of outcome as you expect. So, you’ve tried to actually enhance the public good (participant gives a little laugh) or minimize the public harm, but it’s a very difficult job. (#42, advisory committee)

While the implications of such an oversight in my study are arguably inconsequential in terms of the harm risks, the same would not necessarily be true within the context of NBS.

This check box dimension in my express consent process introduced unanticipated challenges and the potential for error, reflecting – on a small-scale – what a number of my participants identified as potential logistical and implementation fears and concerns regarding express consent for NBS: specifically, that the practice of express consent could inadvertently introduce oversights and mistakes that would not only compromise screening uptake, but parental autonomy as well. In developing my consent form for this study, including in it two questions with “yes” or “no” check boxes, I never expected that many of
my participants would end up completely skipping the check boxes. (I realize that any number of factors could have contributed to this phenomenon from consent form design and layout to sentence structure and phrasing to some other unknown variable.) From these findings, one of course cannot make conclusions about the general effectiveness of a check box approach to express consent. Rather, I disclose this unexpected consent-related phenomenon that occurred in my study to contribute to the list of potential harms worthy of consideration and mitigation in the event that Newborn Screening Ontario, in conjunction with the Ministry of Health and Long-Term Care, decide to pursue – at some point in the future – a check box approach to express consent for NBS on a provincial scale.

By no means should this presentation of my study finding be interpreted as an endorsement of approaches to NBS that do not support parental autonomy and self-determination through an effective approach to consent. Instead, I draw on this finding to support my larger argument that least restrictive alternatives (e.g., express consent) for a medical public health intervention (e.g., NBS) may have unexpected and unintended outcomes that could potentially cause harm: Therefore, approaches to express consent need to be examined and evaluated for effectiveness in terms of their ability to achieve the sought after balance of protecting and promoting both parental autonomy and the public health goals of the program (see Chapters 2 and 8). The findings categorized under this sub-theme “a check box approach to consent”, merely serve as points to consider in future examinations and assessments of least restrictive approaches to NBS moving forward.
Notions of a “simple” consent for newborn screening

As mentioned earlier, many participants did not think that a comprehensive explanation of each of the 28 conditions on the screening panel was ideal, fearing such an approach would neither foster parental autonomy nor NBS uptake. Some participants proposed an alternative to the “all or nothing” (#51, treatment centre) approach to consent for NBS by making an appeal for a “simple” consent process that presents parents with the NBS information basics:

And the more complex you make a consent, I think the more challenging, both in terms of the amount of time it takes to talk about it, answer questions, all of that, and the possibility for refusal. Like, it’s got to be as simple as possible. (#51, treatment centre)

So I just think it’s an excuse when people say it's [explaining NBS is] too complicated. You don't have to get into telling them the names of every single condition, explaining what each of those conditions are. You might have, once in a blue moon, you might have a parent who wants to know, right? So you do have to be able to provide that information, right? Um…but I think most parents, they aren't gonna…they just want to know, you know, oh that these are really serious diseases, you know, I better have my baby checked, right? (#7, legal expert)

[B]asically outline what are the (participant raps the table) outcomes. What are the possible outcomes of this test. That's the main thing that people need to know. You get a result that means you're normal, likely, you get a result that means you’re abnormal in some way, likely you get a result somewhere in the middle which is the most difficult one and…and that might need more testing. (#12, advisory committee)

Participants also expressed the need for the implementation of a conversation process if a parent declines screening to ensure that they fully understand their decision and the possible implications of that decision:

So, this is why if there is dissent then there should be a process to discuss. (#36, treatment centre)
I think there's the general sense [in Ontario] that people will or can opt out. Again, we're talking about a rare event (participant laughs). Maybe one in...I think I'm aware of one or two cases in three years here. And often it's a matter of sitting down with the mom at the time and say, "What are your concerns?" "What are your questions?" Okay, here's the answers. (#15, advisory committee)

Consent forms and processes devoid of “legalese” (#15, advisory committee) were also considered critical to a “simple” consent approach. The perceived legalistic style and nature of express consent processes was explicated by one participant as an impediment to parental knowledge and understanding — “to the detriment of the information to the mom”:

So I think you can see there's that tension between, you know, what we...I think we need ah...to stay at a good level of education and a good uptake of the program versus what some, be they hospitals, institutions, or REBs feel they need, sometimes just to protect themselves against lawsuit [...] (#15, advisory committee)

In short, a “simple” consent approach to NBS that provides parents with the important NBS information basics in a manner that is easy to understand – free from legalese – was considered a possible approach to NBS that could potentially succeed in respecting parental autonomy while also achieving public health goals. In addition, participants insisted that provisions would need to be made to engage in a dialogue with parents who decline screening to ensure that such decisions are based on knowledge and understanding and reflect a truly informed decision.

Decision-aids to support express consent for newborn screening

Finally, a couple of participants put forward the possibility of going “beyond informed consent” (#41, advocate) to presenting parents with a NBS decision-aid that would guide them through the different NBS options according to their values and preferences. The decision-aid was considered a mechanism that “leads people into understanding kind of what
the options are and it helps them to *come up with a decision* as opposed to just putting the information out there and then you have to kind of figure out” (#41, advocate):

I guess the question one would ask is, *at the end of the day* (participant bangs the table), let's say I had a *very good informed model* (participant bangs the table again). And let's say that I even set it up so well that it included not just information, but I could give you some *decision aid*, so it would take you through some Q&A's and it would hopefully lead you to be enabled to analyze the risk and benefits, right? So I've taken you through not only *information* and provided the information in a way that is understandable to you and *also* given you some assistance in terms of sorting through the risks and benefits so you arrive at something. (#41, advocate)

So … I mean the information aid is the perfect example, or decision aids, where people used to think that *all* patients needed was information to make a decision about something. *Clearly*, research has shown that that is *not* the case in decision-making and that if … if that’s all providers do is provide information then they’re not really helping them through a complex decision; whereas if they take into account um…their supports, their values, they’re *much more likely* to help them come to a decision that they will be happy with or satisfied with and have less decision conflict. And you know, that’s something that has morphed over *time*, so research *enabled* that kind of process, so to me the value of research is having us … well there’s a number of values to it generally, but it’s allowing us to make more *informed* decisions about things, that are informed by the people that are affected and the people that are providing, so … and I think that it … I think the other thing is any … any program that’s put in needs to be *evaluated* to determine what are the … the harms and benefits of *that* particular thing and how effective was it. Cuz *again*, it’s very easy to *think* that something you’ve introduced is very effective or is wonderful and everyone loves it, but you could be quite wrong. (#3, advisory committee)

Despite the perceived promise of a decision-aid for NBS, the participants who introduced them as a possibility for NBS in Ontario were very clear that such an intervention would need to be studied and evaluated for effectiveness both in achieving “*truly informed consent*” and achieving the public health goals of the program, namely ensuring that all affected infants are identified and treated:
But *again*, I would argue that anything like that [a decision-aid] should be studied the same way as that other study was done. Because, what impact might it *have* if people stop having newborn screening? Would it affect their decisions? What … like I think information interventions *can be as* ah … well can have the same affect as a drug in that sense … you know you need to follow-up from what happens from that so … so if you put something like that *in place*, would people stop having newborn screening (participant says laughing)? You know, like what would happen (participant still laughing) … it might not … not accomplish what we really want which is informed decision making but *truly* informed … like truly understanding what it is […]. (#3, advisory committee)

This idea of evaluating the decision aid as an “information intervention” is one that I explore more fully in the next chapter (Chapter 8), but I extend this notion of evaluation to express consent for NBS in general.

I interpreted my participants’ discussions around the check box approach to consent, simple consent, and decision aids as perceived least restrictive approaches to NBS that, according to some of my participants, might better succeed in achieving a balance that would support both parental autonomy and public health goals. However, some participants recognized that despite perceptions that these approaches held promise for both self-determination and screening uptake, some participants were adamant that if such approaches are implemented, they would ultimately need to be evaluated for effectiveness in terms of being able to achieve the balance in practice and to ensure that the approach does not interfere with the public health program’s goals. As articulated in Chapter 2, least restrictive alternatives to a public health initiative may not only be ineffective in achieving the public health goals, but can also cause harm (Guttman & Salmon, 2004; Nuffield Council on Bioethics, 2007; Faden & Shebaya, 2010). Therefore, in Chapter 8, drawing on my findings as analyzed and interpreted within a public health ethics lens (see Chapter 2), I put forth a public health ethics framework (Public Health Ontario, 2012) that is useful in assessing the
policy and practice of consent. This framework encourages professionals engaged in public health initiatives to enumerate not only the potential strengths of the intervention under examination, but also to identify the real and potential harms so that they may be preempted prior to implementation on a public health scale.

Chapter Discussion

This chapter reflects my participants’ perspectives on the possibility of an express consent policy and practice for NBS in Ontario as analyzed and interpreted through a public health ethics lens that strives to find a balance between protecting and promoting the public’s health while also respecting the rights and freedoms of individuals. A dominant theme reflected in my data was the perception that an express consent policy and practice for NBS would be potentially harm-causing. I analyzed and interpreted what my participants identified as concerns related to an express consent approach to NBS in Ontario as harms perceived to have implications not only for screening uptake, but also for parental autonomy as well. My data captured a range of perspectives from clear support of express consent to more dismissive attitudes given beliefs that express consent is an inappropriate focus for a public health initiative such as NBS. However, many participants who discussed the issue of express consent for NBS, even if they clearly preferred one approach to the other, revealed their inner-conflict and ambivalence on the matter—appreciating the importance of honouring and supporting parental autonomy, yet fearful that doing so could compromise the public health goals of the program.

Framing informed consent as a least restrictive approach to a public health initiative serves as a reminder that consent may not succeed in realizing its objectives. For as scholars
have argued, however well-intentioned the least restrictive approach might be, it might not succeed in achieving its goals and could introduce unexpected and unintended harms as well (Mcintyre & Petticrew, 2000; Guttman & Solomon, 2004; Nuffield Council on Bioethics, 2007; Tannahill, 2008; Faden & Shebaya, 2010). Public health ethics analyses of least restrictive approaches to public health initiatives, therefore, demand consideration not only of known harms, but of potential harms as well so that they can be addressed in advance of implementation on a public health scale (Kass, 2001; Baum et al., 2007; Tannahill, 2008; Public Health Ontario, 2012; ten Have et al., 2013). Tannahill (2008) argues that to engage in such analyses requires drawing on existing empirical, theoretical, and ethics scholarship. Since public health decisions often need to be made with limited empirical data, drawing on other sources to anticipate potential benefits and harms from public health programs, interventions, policies, and practices is critical (Tannahill, 2008).

Given the exploratory and interpretive nature of my inquiry I looked to existing empirical data that addressed the issue of consent for NBS not only to contextualize my findings within the broader scholarship on this topic, but also to see whether my framing of consent as a potentially harm-causing least restrictive approach to NBS could be supported sufficiently to warrant additional exploration through a targeted inquiry in the future. Empirical research involving parents and HCPs on the topic of NBS generally and consent for NBS in particular suggests that express consent for NBS is not without its challenges. How express consent is implemented and presented to parents has been identified as critical in achieving truly informed, autonomous decisions. Scholars have argued that approaches to consent that induce parents to consent or consent that is sought and obtained in a routine or
perfunctory manner compromise the consent process and/or undermine autonomy

(Hargreaves et al., 2005a; Huang et al., 2005; Newson, 2006; Davis et al., 2006; Parsons et al., 2007; Miller et al., 2010b). Other documented concerns associated with implementing an express consent approach for NBS include the challenge of ensuring knowledge and understanding among parents and HCPs as well as an apparent ambivalence among parents themselves as to whether they want an express consent approach for NBS (Faden et al., 1982; Holtzman et al., 1983; Campbell & Ross, 2003; Hargreaves et al., 2005b; Quinlivan & Suriadi, 2006; Detmar et al., 2007; Moody & Choudhry, 2011).

Existing empirical scholarship on NBS involving parents and HCPs articulates concerns that informed consent practices, both implied and express, are often not succeeding in achieving parental knowledge and understanding (Davis et al., 2006; Detmar et al., 2007; Parsons et al., 2007). Studies have found that the complexity of the information (Liebl et al., 2002; Kemper et al., 2005; Feuchtbaum et al., 2007), the timing of the consent process (Davis et al., 2006; Parsons et al., 2007), sociodemographic factors of parents (which I have interpreted through a public health ethics lens as having implications for social justice) (Lange et al., 2009; Tarini et al., 2010; Araia et al., 2012), and the lack of HCP knowledge coupled with their ability to influence parent decision-making on NBS (Hargreaves et al., 2005b; Parsons et al., 2007; Hayeems et al., 2009) have all been found to interfere with – or hold the potential to interfere with – parent knowledge and understanding within the context of consent for NBS. These study findings arguably lend support to my framing of informed consent (implied and express) as a least restrictive approach to NBS that can be more or less effective. Also, this empirical literature arguably supports my interpretation of consent as
having the potential to cause harm — not only for the program but for autonomy as well.

That certain patterns of concern regarding consent for NBS transcend not only jurisdictions but also study participant groups (e.g., parents, HCPs, NBS program personnel) suggests that these issues should receive focused attention in advance of implementing an express consent approach to screening in Ontario. Among the findings in existing NBS scholarship that may be interpreted as harm include parent perceptions of and reactions to express consent for NBS as potentially too detailed, overwhelming and burdensome, and an approach that may “arouse suspicion and mistrust” in some parents (Detmar et al., 2007; Moody & Choudhry, 2011). An approach to informed consent that overwhelms rather than empowers parents will arguably do little to achieve the goals of consent and risks compromising not only parent autonomy, but also the public health goals of the program.

The feelings of tension and ambivalence participants in my study displayed when contemplating the possibility of express consent for NBS in Ontario are also apparent in other research findings involving study samples with parents and HCPs (Faden et al., 1982; Holtzman et al., 1983; Campbell & Ross, 2003; Hargreaves et al., 2005b; Quinlivan & Suriadi, 2006; Detmar et al., 2007; Kerruish et al., 2008; Moody & Choudhry, 2011; see also Chapter 3). That different groups of study participants articulate a similar challenge of wanting to reconcile two seemingly competing public health ethics principles – namely respecting parental autonomy while also ensuring that the public health goals are met – suggests that a least restrictive approach to NBS that achieves an equilibrium between these two principles would be welcomed. The documented ambivalence regarding consent for NBS among parents, HCPs, and, most recently through my study findings, professionals and
advocates connected to the NBS program itself would appear to support future exploration of the argument that informed consent within a public health context is unique and may need to take a slightly different shape from informed consent within a clinical or research context in order to achieve the aforementioned public health ethics balance. An examination of informed consent through an analytic public health ethics framework is arguably among the first steps of such an inquiry (see Chapter 8).

Chapter Conclusion

In this chapter I presented my participants’ perceptions of express consent for NBS in Ontario. As participants considered the possibility of such a practice their fears surfaced. While participants underscored their concern that express consent for NBS could lead to an increase in parent refusals, mirroring a common concern in the academic literature on NBS, their concerns about express consent extended well beyond parents opting-out. I interpreted their fears and other perspectives in three ways: 1) that the practice of express consent could cause harm to the program, namely identifying affected infants; 2) that the practice of express consent risks devolving into a ritual rather than a true reflection of self-determination and informed, autonomous decision-making, thereby potentially compromising parental autonomy; and 3) the perception that within a public health context such as NBS, express consent might need to be reconceptualized in order to find the balance between reaching population health goals and respecting individual rights and freedoms.

Framing express consent for NBS in these three ways and challenging the practice of express consent to achieve its theoretical ideals is a point of departure for negotiating an approach to express consent for a public health program such as NBS that has a hope of
being effective at both a population and individual level. To aid with this endeavour, in the
next chapter I apply a public health ethics framework to the practice of consent itself to
contribute to the ongoing discussions about whether express consent for NBS is an
appropriate policy and, if it is, how best to integrate it to ensure that it minimizes harms not
only to program uptake but to parental autonomy as well.
Chapter 8. An Applied Public Health Ethics Analysis Of The Practice Of Express Consent

Chapter Overview

Participant perceptions of informed consent for NBS, both implied and express consent, generated a series of perceived challenges to the practice of consent. Specifically, does the practice of informed consent achieve its theoretical goals and objectives and could informed consent introduce unintended outcomes. How individuals charged with advising the Ontario government on such policy issues as consent for NBS perceive the practice of consent will arguably influence, at least to some degree, their recommendations. This case study explored the attitudes towards the policy and practice of consent for NBS from the perspectives of key informants connected and committed to NBS in Ontario, many of whom are responsible, either directly or indirectly, for advising the government on program policies, guidelines, and practices. One of the predominant findings of this research was the perception that the practice of informed consent could introduce harms of its own; not only to the effectiveness of the NBS program’s ability to achieve its public health goals, but also to the effectiveness of the practice of informed consent.

The implied consent approach for NBS in Ontario generated much discussion and debate among participants in my study: some participants perceived implied consent as a relative equivalent to mandatory screening, whereas others clearly identified implied consent as an approach to informed, parental consent. Independent from the disagreement around where implied consent falls on the “autonomy spectrum” (see Appendix E), there was
agreement among those participants who discussed issues related to implied consent that while implied consent is an efficient and effective approach to ensure that all infants are screened, implied consent, as currently practiced in Ontario, was perceived to fall significantly short of maintaining the necessary standard of informed consent. Participants’ perceptions of and attitudes towards the possibility of an express consent approach to NBS in Ontario also highlighted a range of perspectives from complete and forceful objection to express consent even being considered as a topic of discussion given the public health nature of NBS, to a belief that people should always be fully informed and consent expressly to any “genetic” testing. These opposite viewpoints reflect the public health challenge frequently identified in the public health ethics literature: how to find a balance between protecting both the health of the population as well as individual rights and freedoms. Not surprisingly, arguments pertaining to express consent for NBS typically fall clearly along those philosophical orientations as well (Nijsingh, 2007; Ross, 2010). While a few participants’ perspectives could be framed distinctly within these two positions, most participants who discussed their views and attitudes towards consent appreciated the strengths and weaknesses of both positions, enabling me to frame the debate in terms of tensions, ultimately revealing participants’ ambivalence about the approach to informed consent for NBS. Specifically, participants, in many cases, recognized on the one hand the importance of ensuring parents are fully informed and given an opportunity to express their consent explicitly, yet simultaneously articulated their concerns that introducing express consent could compromise the goals of the screening program.

50 Although not all of the conditions on the NBS are genetic in nature, many participants referred to NBS as genetic screening and testing.
To explore, analyze, and interpret these tensions around informed consent for NBS, I used a public health ethics lens. This lens led me to identify four key public health ethics principles that capture the range of perceptions regarding informed consent for NBS: consent as a least restrictive alternative; effectiveness of consent; consent as a challenge to autonomy; and the social justice dimensions of consent. Applying a public health ethics lens for this ethics analysis arguably expands the debate beyond the more traditional arguments that informed consent for a medical intervention is a patient right codified in legislation (*Health Care Consent Act, 1996*; Wildeman & Downie, 2001; College of Physicians and Surgeons of Ontario, 2006). Explicitly framing informed consent for NBS as a least restrictive alternative (see Chapter 2 and Appendix E) that can be more or less effective in achieving not only its public health goals but also, as I have argued, autonomy as well, arguably introduces a lens through which to examine informed consent for NBS in a way that aims to bridge the gap between more liberal and consequentialist framings of the debate.

Increasingly scholars have begun to challenge least restrictive alternatives in the public health context as presumed goods, arguing instead that, like more restrictive approaches to public health, least restrictive approaches to public health initiatives can also cause harm (Guttman & Soloman, 2004; Nuffield Council on Bioethics, 2007; Tannahill, 2008; Faden & Shebaya, 2010). According to some public health ethics scholars, a public health ethics lens of analysis for least restrictive alternatives demands a comprehensive reflection of both the known and potentially hidden harms that may emerge if implemented so that they may be preempted or at least mitigated (Tannahill, 2008; Public Health Ontario, 2012; ten Have *et al.*, 2013). Analytic public health ethics frameworks have been designed
specifically to assist professionals engaged in public health initiatives explore, assess, evaluate, and anticipate the known and unknown harms associated with a given public health program, intervention, policy, or practice (Tannahill, 2008; Public Health Ontario, 2012; ten Have et al., 2013). These frameworks are intended to initiate discussion and debate through the use of guided questions that incorporate key public health ethics principles deemed worthy of consideration within public health contexts to ensure the public health initiative in question is ethically justified.

The emphasis that participants in my study placed on the perceived benefits and harms of informed consent practices for NBS in Ontario led me to consider the ethical justification of informed consent as a least restrictive alternative for NBS using a modified version of Public Health Ontario’s (2012) analytic public health ethics framework, *A Framework for Ethical Conduct of Public Health Initiatives* (see Table 2 p.52). I maintain that using a public health ethics framework such as this within the context of informed consent for NBS creates a third framework through which to discuss and debate perceived tensions related to consent for NBS that extend beyond merely defining consent debates in terms of liberal or consequentialist worldviews. Such an approach to the ethical justification of public health initiatives such as consent attempts to help professionals on both sides of the debate move towards a shared third option that strives to reconcile existing tensions and ambivalence in pursuit of achieving a balance between these two distinct, and at times seemingly irreconcilable, positions.

In this chapter, I provide an applied analysis of express consent for NBS. However, Public Health Ontario’s (2012) framework is arguably a useful lens through which to
examine other least restrictive approaches to NBS as well, including implied consent. I decided to focus on express consent for this particular analysis in an effort to respond to the growing body of scholarship calling for a reevaluation of mandatory and implied consent approaches to expanded NBS in favour of express parental consent as a result of the expanding nature of the NBS programs and the ethical issues such expansion introduces (Wildeman & Downie, 2001; Provincial Advisory Committee on New Predictive Genetic Technologies, 2001; Ross, 2010). As NBS programs in Ontario and elsewhere consider the possibility of such a transition, Public Health Ontario’s (2012) comprehensive public health ethics framework could be a useful tool to help professionals charged with decision-making (or those serving in advisory capacities) consider the ethical justification of express consent for a public health intervention by examining the benefits and potential burdens of implementing such a least restrictive approach. Also, this applied analysis will focus predominantly on the ethical justification of express parental consent for the initial newborn screen. The question of whether express parental consent should be obtained prior to the initial newborn screen is arguably foundational to the overarching consent strategy for the many other dimensions of expanded NBS, such as long-term storage of and future uses (including research) for dried newborn blood spots. The approach to consent adopted for the initial screen will have implications for how best to design and implement informed consent processes for the other aspects of the program. Although this applied analysis focuses mainly on the ethical justification for express consent for the initial screen, where applicable I include my analysis and interpretation of participants’ perceptions of the perceived
advantages and disadvantages of express consent for storage of and future uses for dried blood spots (see Chapter 7).51

I divided this chapter into two parts. In Part I of this chapter I defend the framing of express consent as a least restrictive approach to NBS that merits ethical justification from a public health ethics perspective. Then, in Part II I apply a modified version of Public Health Ontario’s (2012) *A Framework for Ethical Conduct of Public Health Initiatives* (described fully in Chapter 2) to my findings on express consent for NBS in Ontario. The goal of this applied public health ethics analysis is to ask questions of the practice of express consent for NBS through a lens not typically used to examine consent. The responses I provide for these guiding questions serve as points of departure for future discussion and elaboration and are drawn not only from my findings, but also the findings from existing literature — an approach to public health ethics analysis consistent with those of other scholars, whereby empirical data must be supplemented with theoretical and ethics scholarship (Tannahill, 2008). Public Health Ontario’s framework facilitates a comprehensive analysis of benefits and harms connected to express consent for NBS and surfaces potential harms that arguably should be addressed in advance of a decision to implement express consent for NBS.

*Part I: Informed Consent as a Least Restrictive Approach to Newborn Screening that Warrants a Public Health Ethics Analysis*

Informed consent in healthcare is an ethical concept that promotes respect for persons and an individual’s capacity to determine his or her course in life without external influence.

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51 The ethical issues related to the storage of and future uses for dried newborn blood spots, among them consent, are significant and worthy of an independent, focused inquiry (see Chapter 9). The exploratory nature of my study renders such a targeted, comprehensive focus on storage and future uses outside the scope of this analysis.
or coercion (Beauchamp & Faden, 1995; Paterick et al., 2008). The process of informed consent demands more than mere disclosure of information: patients (or their surrogate decision-makers) and potential research subjects must have a full understanding of the information presented to them and they must be free to make decisions voluntarily without coercion (Nuremberg Code, 1947; Beauchamp & Faden, 1995; O’Neill 2004a; Patrick et al., 2008). As articulated in the introductory chapter of this thesis, informed consent in Ontario may be either implied or express (Health Care Consent Act, 1996).

According to the Health Care Consent Act (1996), the informed consent requirements for implied and express consent are the same. In the context of implied consent, consent is assumed, and within the context of express consent policies, consent must be verbalized or written. Implied consent is an approach to consent that has been challenged within the context of NBS, questioning whether HCPs are able to presume fully informed consent from the absence of patient or surrogate decision-maker refusals (Wildeman & Downie, 2001). Beyond debates around implied consent for NBS, the College of Physicians and Surgeons of Ontario (2006) generally discourages the practice of implied consent for medical treatment in favour of express consent. Despite perceived limitations of implied consent, the practice of express consent has been called into question as well by scholars examining clinical ethics (Dawson, 2005; Crepeau et al., 2011), research ethics (Flory & Emanuel, 2004; Dawson, 2005; Dawson, 2006); and public health ethics (O’Neill, 2004b; Nijsingh, 2007). Not surprisingly, such challenges are increasingly debated within the context of NBS programs as well (Faden et al., 1982; Holtzman et al., 1983; Hargreaves et al., 2005a; Hargreaves et al., 2005b; Nijsingh, 2007; Nicholls, 2010). As Ontario reflects on its own approach to informed...
consent for NBS (Born Ontario, 2013a), Public Health Ontario’s (2012) public health ethics framework may be a useful tool to facilitate discussion and debate moving forward.

Empirical studies examining express consent in clinical and research domains have found that patients and research subjects may consent to healthcare interventions and/or procedures or clinical research initiatives without completely comprehending what they consented to (Flory & Emanuel, 2004; Tait et al., 2005; Dawson, 2005; Crepeau et al., 2011), a reality which arguably undermines the practice of consent. Moreover, such studies have found that while significant gaps in comprehension are demonstrated across study participant demographics, participants with less education were found to comprehend less than those participants with higher education levels (Tait et al., 2005; Crepeau et al., 2011). In studies where sociodemographic variables contribute to discrepancies regarding knowledge and comprehension, one might argue that the autonomy of some patients and research subjects is compromised more than others, and therefore raises questions of social justice.

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52 “The Maternal/Child Screening Committee will make recommendations to the Provincial Council on Maternal and Child Health (PCMCH) and to BORN Ontario to ensure that:

• the scope and operation of prenatal, newborn and childhood screening in Ontario meet the changing needs of the people of the province;

• consistent general principles of screening, including standards of implementation and evaluation, are applied to all aspects of prenatal, newborn and childhood screening, including, but not limited to, the recommendations of the Prenatal Screening Committee and the Neonatal Screening Committee. Examples include, but are not limited to, decisions relating to the disclosure of adventitious information (unanticipated findings made in the course of screening), consent, storage of samples, access to stored samples, utility of new screening technologies, etc.; […]” (Born Ontario, 2013a).

53 The purpose of alluding to studies exploring the limitations of obtaining express consent in clinical and research contexts is to highlight a shared challenge across the various medical contexts. Although express consent for NBS would be a public health initiative implemented on a province-wide scale, the actual consent practice would be experienced by individual parents. Therefore, being aware of the way in which autonomy may be compromised in other domains is critical. Express consent that is compromised and undermined is problematic in any medical context. However, failed consent at a public health level arguably could, according to my participants, have implications for infants, parents, and society further underscoring the importance of ensuring, to the degree possible, that an express consent process as effective is as possible prior to universal implementation.
While poorly designed, implemented, and executed consent processes within clinical and research contexts can have significant ramifications for the patient or research subject — not an inconsequential outcome — an ineffective consent process at a public health level can potentially effect the well-being of populations. While respect for individual liberty and self-determination through autonomous decision-making means that HCPs must respect decisions they might ultimately disagree with (Wildeman & Downie, 2001), if those decisions are made out of patient or surrogate decision-maker misunderstanding due to a poor design and/or implementation of express consent, the extent to which those individuals made autonomous decisions can arguably be challenged.

The documented challenges of achieving informed decisions through express consent practices have led to research initiatives that strive to improve the effectiveness of informed consent approaches in the clinical and research domains to ensure, to the extent possible, that consent practice meets its theoretical objectives, namely obtaining truly informed, autonomous decisions (Jimison et al., 1998; Flory & Emanuel, 2004; Tait et al., 2005). Express consent processes have been analyzed and consent forms have been scrutinized in a range of healthcare contexts in an effort to improve the process of consent and narrow the gap between informed consent in theory and in practice (Tait et al., 2005; Dawson, 2005). Consent forms in various studies have been deconstructed for reading level, comprehensibility, and visual ease and appeal (e.g., font size, white space, pictographs) (Tait et al., 2005; Kang et al., 2009). Some studies have also experimented with the effectiveness of accompanying the consent discussion and forms with technological aids such as computer programs and videos (Jimison et al., 1998; Flory & Emanuel, 2004). Other scholars have
conducted studies to see whether alternative approaches to obtaining informed consent such as using computers rather than forms improve comprehension and knowledge (Jimison et al., 1998; Flory & Emanuel, 2004). These studies write about “modifying” and “simplifying” the practice of consent to improve patient and/or research subject knowledge and understanding. That there are potentially better and worse ways of operationalizing express consent in the pursuit of protecting and preserving individual autonomy – and that those approaches can be tested and evaluated according to specific criteria – further supports framing informed consent practices as least restrictive approaches to a medical public health intervention in need of applied public health ethics analyses.

My argument, in short, is that express consent, as an ethical concept, has clear objectives that are not always met in practice. Research shows that efforts are continuously made to improve the practice of express consent to ensure, to the extent possible, that express consent meets its objectives. Dating back to the Nuremberg Code (1947), the requirements necessary to obtain informed consent – referred to as “voluntary consent” – from potential research subjects were well delineated and evolved to form the core requirements of informed consent in clinical care context (Faden & Beauchamp, 1986; see also Chapter 1). These expectations for informed consent are very clear and include that patients and research subjects make autonomous decisions that are rooted in knowledge and understanding and that are free of coercion (Nuremberg Code, 1947; Faden & Beauchamp, 1986; Health Care Consent Act, 1996). Although clear in theory, the question is whether the objectives of informed consent are achieved in practice to the extent that they enable and protect individual autonomy with respect to healthcare decisions. Express consent is meant to respect persons:
autonomous, rational agents capable of self-governance. However, every time patients, surrogate decision-makers, or research subject candidates give their consent without knowledge and understanding, one might argue, as some scholars have, that the goals and objectives of consent have not been met and patient autonomy is compromised (Hargreaves et al., 2005a; Huang et al., 2005; Newson, 2006; Kerruish et al., 2008). Within a public health context such as NBS, compromised express consent practices could have implications not only for parental autonomy, but also the public health goals and objectives. Within a public health context, compromised expressed consent practices undermine the “consent” obtained and hold the potential to cause harm not only to infants, but to parents and to society. A poorly designed and/or executed consent process risks inflicting not only “dignitary harms”\(^{54}\), but also physical and psychological harms.

Some scholars might argue that the real and/or anticipated challenges of express consent for NBS identified not only by my participants, but also other researchers publishing on the topic of express consent in various healthcare contexts, is a failure of the program charged with implementing consent or a clear sign that the HCPs are derelict in their fiduciary responsibilities to their patients and/or potential research subjects. However, that such challenges and failures of express consent are pervasive across the clinical, research, and public health ethics communities suggests that there may also be challenges embedded within the practice of express consent itself. Framing informed consent as a least restrictive approach to a public health program enables a public health ethics analysis of the practice of express consent for NBS and an opportunity to identify the existing and potential benefits

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\(^{54}\) Dignitary harms are defined as a failure “to show respect for persons (individuals or communities), even where no consequential harm or injury has occurred, such as through privacy breaches or use of information without consent” (Public Health Ontario, 2012: 14).
and harms of the practice in order to minimize potential harms prior to implementation on a public health scale. In addition, a public health ethics analysis of the practice of express consent works towards achieving a middle ground between liberal and consequentialist conceptualizations of healthcare in a community.

Having explained my rationale behind express consent as a least restrictive approach to NBS that could benefit from an applied public health ethics analysis, I will now apply my study findings on perceptions of the practice of express consent for NBS to a modified version of Public Health Ontario’s (2012) public health ethics framework (see Chapter 2 for more details). Decision-making in public health contexts must often occur with limited empirical evidence (Tannahill, 2008). Therefore, Tannahill (2008) encourages considering scholarship that extends beyond empirical data to include theoretical and ethical scholarship as well prior to implementing a public health program, policy, intervention, or practice. In this applied public health ethics analysis I will also draw on existing empirical, theoretical, and ethics literature on informed consent and NBS to initiate preliminary responses arguably worthy of future exploration to each of the ten questions in the framework.

Part II: A Framework for Ethical Conduct of Public Health Initiatives Applied to Express Consent

While the ethical concept of consent is rooted in liberal understandings of self-governance, how informed consent is operationalized and practiced is of the utmost importance to ensure, to the extent possible, that individuals faced with health decisions – and in the case of NBS public health decisions – are actually exercising their autonomy in a truly informed manner. Framing consent as a least restrictive alternative within a broader
public health program such as NBS – as an “initiative” or “information intervention” in its own right with better or worse effectiveness – facilitates a comprehensive analysis of the practice of consent that arguably provides the insights necessary to work towards narrowing the gap between the two extremes of protecting the rights and freedoms of individuals on the one hand and the health of the population as a whole on the other. By applying Public Health Ontario’s (2012) framework to the practice of consent itself rather than merely assessing the issue of informed consent within the broader discussion of NBS forces a line of inquiry that challenges the taken-for-granted assumptions embedded in consent. By using such an analytic tool I hope that my findings from this inquiry might contribute to the dialogue in Ontario and elsewhere about consent for NBS and consent for public health programs and initiatives more broadly.

Applying the Framework’s 10 Questions to the Practice of Express Consent for Newborn Screening

1. What are the objectives of the initiative? How are they linked to potential improvements in public health?

Proponents of express consent for NBS argue that the public health risk posed by rare conditions identified through NBS does not justify infringing on a parent’s right to make health care decisions for their children: in the case of NBS, the initial decision in question is whether to have their infant screened or not (Newson 2006; Ross, 2010). The objectives of implementing express consent would be to encourage meaningful parental autonomy, foster informed decision-making with respect to NBS, and, as a result of the transparency, fortify
trust in Newborn Screening Ontario and any future initiatives that may arise within the context of NBS.\textsuperscript{55}

In public health, emphasis is often placed on the extent to which a given intervention reduces morbidity or mortality (Beauchamp & Steinbock, 1999; Kass, 2001; Childress \textit{et al.}, 2002; Benatar, 2003; Jennings, 2003; Baylis \textit{et al.}, 2008; Dawson, 2009; Dawson, 2010). Kass (2001) also makes explicit that decreasing psychological morbidity is a legitimate end for a public health intervention as well. Preempting or mitigating dignitary harms is also among the ethical considerations within the context of a public health initiative (Public Health Ontario, 2012). In considering the practice of express consent for NBS from a theoretical and ethical perspective, the Ontario government might contemplate implementing express parental consent for NBS because it arguably epitomizes the least restrictive approach to achieving public health goals and objectives (see Appendix E)—an approach to public health initiatives that is strongly encouraged in public health ethics scholarship (Upshur, 2002; Childress \textit{et al.}, 2002; see Chapter 2). Use of least restrictive alternatives for public health initiatives is also believed to incur public trust, an important component of any public health program (Thompson \textit{et al.}, 2006; Tannahill, 2008; Public Health Ontario, 2012). Moreover, seeking express parental consent for NBS acknowledges parents as the lawful surrogate healthcare decision-makers for their children and express consent would honour this parental right directly rather than through implied consent which has been challenged as an approach to informed consent not only for NBS but for clinical interventions generally (Wildeman & Downie, 2001; Provincial Advisory Committee on New

\textsuperscript{55} Transparency is a critical principle in public health ethics and essential for public health programs trying to instill public trust in a given public health initiative (Thompson \textit{et al.}, 2006; Tannahill, 2008; Public Health Ontario, 2012).
Predictive Genetic Technologies, 2001; College of Physicians and Surgeons of Ontario, 2006). Introducing express consent for expanded NBS and its many facets could minimize or potentially remove many of the ethical dilemmas currently debated regarding the dimensions of NBS that extend beyond immediate infant health concerns, such as the ethical justification of storage of and future uses for dried newborn blood spots. Finally, adopting an express consent approach for NBS could also be interpreted as an effort to cultivate the well-being of a population by fostering self-determination, respect, and self respect among its community members (Powers & Faden, 2006).

Other possible benefits of implementing an express consent approach for NBS in Ontario could include a positive impact on the psychological and emotional well-being of parents whose infants screen positive for one of the conditions on the screening panel. A number of my participants, particularly those who worked in the treatment centres, discussed their perceptions of the differences in parental anxiety between parents who were aware that NBS had occurred (lower anxiety) and those parents who were not aware (higher anxiety). Making a distinction between informed parents and consent is important, as the former can arguably occur without the latter. However, given that parent knowledge and understanding are critical to any informed consent initiative, effective parent education would be a core component of an express consent process that strives to honour parental autonomy within the context of a medical public health intervention.

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56 As the advisory committee continues to examine least restrictive alternatives for NBS that strive to achieve a balance between accomplishing the public health goals and objectives while also protecting individual rights and freedoms, improved parent education independent from express consent – tied perhaps to another least restrictive autonomy-preserving approach to NBS – is also an initiative within the context of NBS that could benefit from a similar assessment of the strengths and weaknesses using this analytic public health ethics framework.
Finally, the third possible public health benefit of an express consent approach could be to the overall efficiency and effectiveness of the NBS program itself. Specifically, a number of participants spoke about how often the NBS requisition forms are filled out with incomplete contact information.\textsuperscript{57} This concern was reiterated within the context of the round table discussion I attended at the Newborn Screening Ontario Clinical Follow-Up Symposium in 2010, whereby attendees discussed the challenges and implications for those infants who screen positive and are lost to follow-up. Although an express consent approach to NBS could potentially mitigate this outcome through the explicit education component directed at each parent, other approaches to parent education could potentially serve to address this occurrence. However, that another least restrictive alternative approach could, for example, conceivably influence parents’ decisions to include multiple contact numbers on the requisition form does not diminish the potential public health benefit of achieving such ends through an express consent approach.

However, as will be explored in greater detail throughout this analysis, participant perspectives – some of which are corroborated in the literature\textsuperscript{58} – suggest that express consent for NBS could have negative health implications as well, not only for the infant, but also the parents, the population as a whole, and even the concept of informed consent. As one considers the potential public health benefits of an express consent approach to NBS, they must be weighed against the potential harms (Kass, 2001; Public Health Ontario, 2012). If the advisory committee recommends pursuing an express consent approach for expanded

\textsuperscript{57} It is important to note that with respect to incomplete information on the requisition cards, one might question whether this is a result of parental or HCP oversight—a possible area for future investigation.

\textsuperscript{58} See Chapters 3 and 7 as well.
NBS in Ontario, efforts should be made to identify known and potential harms that could emerge as a result of implementing such a practice. The questions put forth in this public health ethics framework are designed to generate a comprehensive picture of the strengths and weaknesses of this least restrictive alternative so that the potential burdens can be addressed in advance of widespread implementation to mitigate the potential for unforeseen harms.

2. **Can the objectives be achieved using the proposed approach?**

Express consent evoked certain assumptions among my participants. As explicated more fully in Chapter 7, perceptions of what express consent for NBS would look like varied. Some participants felt that within the context of NBS informed decision-making could be achieved following a general discussion around the reasons for NBS, the kinds of conditions on the NBS panel, an explanation of the process that will occur following the heel prick, as well as the basic health outcomes that could be revealed in the event that follow-up testing is indicated (e.g., false positive, true positive, or inconclusive/ambiguous result). Other participants maintained that express consent would require a detailed explanation of each of the 28 conditions on the panel – replete with benefits and risks of screening for each – as well as existing treatment options. Participants who felt that this comprehensive approach was imperative to honour the concept of consent also felt that such an approach would be untenable from an implementation standpoint and potentially ineffective in terms of generating informed decisions.

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59 Modified question: The original question from Public Health Ontario’s framework asked, “Can the objectives be achieved using the proposed methods?” Given the research specific target of this framework, “methods” refer explicitly to study design. I modified this question and changed “method” to “approach” to reflect the perceived need of my participants to find new ways of obtaining express consent.
Participants also debated whether written consent would be necessary or if verbal consent would be sufficient. Participants perceived both options as introducing significant logistical challenges with ethical implications. In particular, participants introduced tracking a documented express consent process on a province-wide scale as a hurdle. My findings also revealed that relying on verbal consent for confirmation could pose a unique challenge in light of the fact that parents could confuse prenatal and newborn screening, which could undermine their decision making, their autonomy, and the consent process itself. The tracking dimension of express consent and the potential harms introduced as a result of implementing such an approach were perceived as being further complicated by introducing consent for the storage of and future uses for the dried blood spots. Specifically, some participants felt that the more dimensions of NBS for which parents could consent, the increased likelihood of introducing errors.

Since NBS has expanded to include more conditions on the screening panel, carrier status disclosure, and storage of and research on dried blood spots (Newborn Screening Ontario, n.d.a; n.d.f), some participants shared with me the different ways in which they thought express consent could be implemented for NBS. For example, some participants mentioned the possibility of introducing check boxes on the actual NBS requisition form; creating a two-tiered screening program with some conditions on the panel mandated (e.g., PKU, CH, and MCAD) and others voluntary; offering a “simple consent”; and the possibility of developing decision-aids in an effort towards achieving truly informed decisions. On the surface, such strategies were considered a means through which to improve consent mechanisms for parents and present them with concrete choices given the expanded nature of
the NBS program. However, although these possible least restrictive approaches were seen as having advantages, participants who discussed these possibilities also recognized that they introduced ethical challenges of their own as well. For instance, some participants questioned whether a check box model would necessarily ensure informed decisions. Regarding the two-tiered approach, some participants noted that what can be separated on paper may not be able to be separated in the lab, thereby raising the ethical question of what to do with incidental results. A “simple consent” approach to NBS was considered by some participants to undermine the construct of consent. Finally, regarding the possibility of introducing a decision-aid, a couple of participants felt such an intervention would require evaluation to establish that the goals of the screening program are not compromised. That some participants introduced the possibility of alternative approaches to express consent (and identified the perceived potential strengths and weaknesses of each) suggests their commitment to informing parents and respecting individual liberties. However, they were simultaneously skeptical that express, written consent – or the aforementioned alternatives – would be effective in achieving informed decision-making.

Existing scholarship on the topic of express consent for NBS involving parents (as explained in greater detail in Chapter 3) suggests that my participants may be justified in their reservations about express consent for NBS as currently implemented in some jurisdictions. A couple of studies addressing consent for NBS from the parent perspective found that many parents do not want lots of details — especially not right before the heel prick (Hargreaves et al., 2005b; Davis et al., 2006). While parents in these studies felt that more comprehensive information should be available for parents who want more detailed
information, some parents articulated their concerns that comprehensive information about
NBS from the outset would be overwhelming, lead to information overload, and/or render
them incapable of evaluating the information in order to make an informed decision (Detmar
et al., 2007; Moody & Choudhry, 2011). Some parents in the study by Detmar et al. (2007)
endorsed choice for NBS but confessed that they were glad they did not need to make a
choice:

It was, in particular, the respondents who advocated freedom of choice who also stated
that this decision would be very stressful […] Many participants said that they were
happy they did not have to make the decision, as demonstrated by statements, such as
‘I’m glad I don’t have to decide any more’. (p.241)

Other study findings that challenge express consent and its ability to foster truly
informed decisions point to the high literacy levels of consent forms and NBS pamphlets and
other informational materials (Holtzman et al., 1983; Hargreaves et al., 2005a; Hargreaves et
al., 2005b; Arnold et al., 2006). Such findings, I would argue, work collectively to challenge
the extent to which informed consent, as currently operationalized in certain jurisdictions,
promotes autonomous decision-making or merely the illusion of choice.60 Some studies,
however, found that express consent can succeed in improving parents’ comprehension of
NBS (Holtzman et al., 1983). Nicholls (2010) suggests that many of the evaluating
mechanisms used to assess knowledge and comprehension focus disproportionately on recall
ability, which may not actually determine understanding.

My participants’ concerns regarding what I have interpreted as the effectiveness,
autonomy, and social justice dimensions of express consent underscore the argument that

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60 High literacy levels of information pamphlets have implications in jurisdictions that have implemented
mandatory screening programs with opt-out provisions as well as those programs that use an implied consent
approaches.
express consent should not be implemented without careful consideration of the potential unanticipated eventualities of the practice of consent itself. In addition, one could argue that what is considered an effective approach to consent within a clinical health context might not translate as well within a public health context such as NBS. Promoting respect for persons through self-determination and autonomous decision-making within a public health context may require re-conceptualizing what express consent for NBS looks like in practice in order to ensure that the goals and objectives of the concept of consent itself are achieved while also working to protect the public health objectives.

3. Who are the expected beneficiaries of the program, intervention, or practice? 

“Who benefits?” is a key question at the heart of any public health initiative. Public Health Ontario’s (2012) *A Framework for the Ethical Conduct of Public Health Initiatives* stresses the interconnectedness of the social fabric of any given community: what helps an individual by extension helps the community and vice versa. In considering the implementation of express consent for NBS, the primary beneficiaries would be the parents of the infants. If the practice of express consent achieves its objectives in terms of supporting “true” rather than illusory parental autonomy, then parents will, ideally, make

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61 That express consent has been challenged in terms of its effectiveness as a medium through which informed decisions are made in the one-on-one encounter in clinical and research settings (Tait et al., 2005; Kang et al., 2009; Crepeau et al., 2011) further supports an argument for reconceptualizing the practice of express consent for a public health initiative.

62 Although an in-depth exploration of what a re-conceptualization of consent for public health programs and interventions might look like is outside the scope of this dissertation, my interpretation of the findings in relation to existing research on consent has led to this possible conclusion, which may be worthy of future debate.

63 The original question 3 in this framework reads, “Who are the expected beneficiaries of the knowledge gained or other benefits?” (Public Health Ontario, 2012: 13). I modified the question to be more suitable for a public health ethics analysis of public health interventions, programs, and practices.
informed decisions that are in line with their values and what they believe to be in the best interest of their infants’ health.

In addition, some participants in my study identified express consent for NBS as an approach that would serve to minimize parental anxiety – an example of psychological morbidity – in the event they receive a call that their infant has screened positive for a condition on the screening panel: a perceived benefit of being sufficiently informed and aware of the possibility of false positives. That express consent could minimize parental anxiety is also articulated in the literature as an argument in favour of such an approach (Sorenson et al., 1984; Hewlett & Waisbren, 2006; Gurian et al., 2006; Schmidt et al., 2012). Connected to the challenge of false positives, some studies have found that parents whose infants receive a false-positive diagnosis can be susceptible to increased anxiety and create what has been described as “vulnerable child syndrome” — never fully accepting that their child is not sick (Clayton, 1999; Schoen et al., 2002; Waisbren et al., 2003; Campbell & Ross, 2003; Kerruish & Robertson, 2005; Clayton, 2005). Such a reaction to a “side effect” of NBS is arguably not good for the parents’ welfare and may negatively impact child welfare as well. Such outcomes could potentially impact society as well, particularly as trips to the emergency room are costly and use limited resources that could otherwise be spent tending to those who are really ill (Waisbren et al., 2003). Improved education about false positives through an express consent process could conceivably serve to minimize such potential outcomes given that parents would, ideally, better understand NBS and what it means to receive a false positive.⁶⁴ Improved psychological and emotional health among

⁶⁴ Improved education regarding false positives is of course not limited to an express consent approach and could be incorporated into the education foundation of other least restrictive approaches, such as implied consent.
parents introduces the possibility of health benefits for their infants and, as a result, society could arguably benefit by extension.

4. What are the burdens and potential harms associated with the proposed initiative? Who bears them?

As outlined extensively in Chapter 7, participants perceived many potential harms associated with an express consent policy for NBS. The potential burdens of express consent would be borne, according to some of my participants, by different members of the community. In addition, a poorly implemented express consent process was also considered a potential harm to the concept of consent and parental autonomy in particular. If express consent is not designed and implemented in a way that enables parents to make truly informed decisions about NBS, then express consent becomes a ritual: a mere symbolic representation of individual liberties and freedoms that, in reality, actually compromises parental autonomy and self-determination. If parents are making decisions on behalf of their infants without grasping fully the NBS information presented to them, they might make a decision they would not otherwise make. The very nature of respecting individual liberty requires abiding by decisions that some may disagree with – including declining NBS on behalf of their infant. However, if parents make a decision to decline screening as a result of misinformation, misunderstanding, or information overload, not only is parental autonomy undermined, their unscreened infants are, according to my participants, left vulnerable — a harm at the forefront of many participants’ concerns when considering who would bear the burden of express consent.
The literature also identifies burdens connected to express consent for NBS. Specifically, if an approach to express consent requires written materials (e.g., pamphlets and consent forms) that are designed in such a way that they are difficult to understand, overwhelming with details, and/or presented at an inopportune time (e.g. following labour and delivery) the practice of consent may be argued as burdensome. Public Health Ontario’s (2012) framework articulates that “[u]nnecessarily burdening a participant is disrespectful, and may harm the goals of an initiative by discouraging participation or prompting early withdrawal” (p. 13). Within the context of express consent, the same outcome can arise: parents could feel overwhelmed by the nature of consent and decline screening without giving it the necessary consideration. Connected to the conceptualization of “consent as a burden” is the timing of such a process. For example, in some empirical studies that examined parent attitudes towards consent for NBS, researchers found that many parents were adamant that seeking express consent post-delivery was less than ideal (Davis et al., 2006; Detmar et al., 2007; Parsons et al., 2007; Moody & Choudhry, 2011). Many described their postpartum experiences and were insistent that decision-making under such circumstances could be compromised (Davis et al., 2006; Detmar et al., 2007; Parsons et al., 2007; Moody & Choudhry, 2011). NBS decisions made under such conditions could arguably be deemed a result of circumstance rather than autonomy.

If express consent is implemented for NBS in Ontario and ultimately leads to a significant number of parents declining screening and, as a result, an increase in the number of infants diagnosed either postmortem or too late to intervene with treatment, a number of participants in my study felt that irreparable harm would have been done: harm borne in the
gravest way by the infant. However, society was also considered among those who would bear a significant financial and moral burden if affected children could have been treated had their condition been diagnosed through NBS. In the event that a parent declines screening and then their child is later diagnosed with one of the conditions that could have been detected at birth, the possibility remains that parents could conceivably sue Newborn Screening Ontario and/or the Ontario government on the grounds that they did not understand what they were declining. Similar fears were also associated with the perceived harms associated with the mechanics of an express consent process regarding the logistics and implementation of such an approach at a provincial level.

The issue of express consent for the storage of and future uses for dried newborn blood spots introduced additional concerns for some participants. Of the participants in my study who discussed these issues, most were adamant that express consent was essential for storage and future uses that extend beyond infant health, program evaluation and/or quality assurance. However, participants were concerned that introducing the expanded dimensions of the NBS program could serve as a deterrent for parents. Specifically participants feared that parents would be worried about the storage and/or research components of NBS and, therefore, in an effort to prevent any future research they would decline the screen altogether. Key questions, therefore, become whether seeking express consent for storage and/or future uses impedes screening uptake and, if it does, should the storage of and future uses for dried newborn blood spots continue within the context of newborn screening. These are critically
important empirical questions, yet they fall solidly outside the scope of this exploratory study.\textsuperscript{65}

Once the burdens and harms have been identified, “[w]here possible, an effort must be made to mitigate or minimize risks and burdens, balancing this against any loss in potential benefit” (Public Health Ontario, 2012: 20). Many of the perceived harms associated with express consent challenge the extent to which express consent actually does what it is supposed to do: facilitate truly informed, autonomous choices. Challenges to the effectiveness of express consent are not new. Scholars have been highlighting challenges to express consent for decades, in particular underscoring skepticism around patients’ abilities to comprehend fully the information presented to them as well as their ability to retain that information over time (Cassileth \textit{et al.}, 1980; Penman \textit{et al.}, 1984; Joffe \textit{et al.}, 2001). In addition, such challenges to express consent are mirrored in some of the existing NBS scholarship reporting study findings involving parents and HCPs (Hargreaves \textit{et al.}, 2005a; Davis \textit{et al.}, 2006; Moody & Choudhry, 2011).

Framing express consent as a least restrictive alternative that has the potential to introduce burdens and cause harm facilitates a comprehensive analysis of such possibilities and, in so doing, creates a list of possible harms that need to be considered and/or addressed in advance of implementing the approach province-wide. The public health nature of NBS creates a tension between balancing the rights of the individual against broader population health objectives. Therefore, identifying the perceived burdens and harms associated with express consent offers an opportunity to reassess and/or reconceptualize the way that consent

\textsuperscript{65} An empirical study designed to assess whether parents are able to trust Newborn Screening Ontario that their choices will be respected if they consent to the screen but wish to have the blood spots destroyed shortly thereafter would be informative.
is designed and implemented for public health initiatives in an effort to ensure that the practice achieves its goals of promoting autonomous decision-making while also promoting population health.

5. **Are burdens and potential harms justified in light of the potential benefits to individuals and/or society?**

   Although not all conditions on the NBS panel are genetic conditions (e.g., congenital hypothyroidism), a number of participants in my study discussed NBS in relation to genetics and genetic literacy. Specifically, some participants disclosed their perception that as the genomic era continues to unfold, lay individuals and HCPs are going to have to acquire a better grasp of genetics and what that means for their health and the health of their families or, in the case of HCPs, their patients. As NBS continues to expand in terms of the conditions for which it screens (e.g., Severe Combined Immune Deficiency (SCID) is scheduled to be added to Ontario’s NBS later this year (Newborn Screening Ontario, n.d.h)), the number of incidental findings it generates, the long-term storage of dried newborn blood spots, and the potential usefulness of the blood spots for research, the arguments supporting mandatory screening and implied consent for NBS will become increasingly difficult to defend (Ross, 2010). However, an ill-conceived and/or poorly implemented consent process can arguably cause harm as well and, therefore, consideration should be paid to minimize potential harms from the practice of consent as a least restrictive alternative.

   Points to consider during such an assessment of express consent could include the relational nature of individuals and communities and the interconnectedness of the benefits

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66 The original question from the Public Health Ontario (2012) framework read, “Are burdens and potential harms justified in light of the potential benefits to participants and/or society?” (p.14). I changed “participants” to “individuals” in order to make it more appropriate for public health interventions.
and harms of express consent for NBS. Moreover, the fact that different kinds of harms are potentially borne by infants, parents, and society may add additional challenges to decision-making on the subject. For example, the dignitary harms of not implementing an express consent policy and practice for NBS are borne by parents, as are the psychological and emotional harms should parents with little knowledge of NBS receive a bewildering and anxiety-provoking call informing them that their child screened positive for a condition on the NBS panel. On the other hand, if an express consent process is implemented but has not been constructed in a way to foster truly informed decision-making but instead merely provides the illusion of choice, not only does this arguably compromise and undermine parental autonomy, but also infants who remain unscreened due to mistakes, misinformation, or misunderstanding may be vulnerable to irreversible morbidity or even mortality.

Framing express consent as a least restrictive alternative to NBS that has the possibility of causing harm as a result of a multitude of factors ranging from the mechanics of implementing a province-wide express consent process to the actual information disclosure dimension of express consent (e.g., how much information to disclose, how to disclose it, when to disclose it, the knowledge base of the HCP disclosing it, and the effectiveness of such disclosure for autonomy and public health) arguably requires time to address all the known and hidden challenges associated with such an approach to ensure a successful approach that achieves the public health ethics balance of meeting public health goals and truly respecting and supporting parental autonomy. Consequently, I would argue that if Ontario decides to proceed with the design and implementation of an express consent approach for NBS, continued efforts should be made to enhance parent and HCP knowledge
and understanding of NBS to support the future express consent process and, in so doing, potentially improve the current implied consent approach. Particular emphasis should be given to the storage and research dimensions of the program, as this information is absent from the informational materials given to parents (see Appendices F-G). Many participants were adamant that independent from an express consent approach for NBS improved NBS education was deemed imperative for both parents and HCPs.67 HCPs were considered by a number of my participants to be a potential barrier to express consent given what some of my participants perceived to be a lack of knowledge regarding NBS. Once Newborn Screening Ontario can feel confident that HCPs and parents have an improved grasp of NBS, at least one important part of the foundation will have been laid for the possibility of introducing meaningful express parental consent.

Technology continues to advance and a few participants in my study alluded to genome sequencing as part of the future for NBS programs — a future trajectory forecasted in the NBS literature as well (Green et al., 2004; Alexander & van Dyck, 2006; The President’s Council on Bioethics, 2008). Such possibilities elicit additional concerns regarding the receptiveness of parents to such continued expansion; however, existing research shows that parents are perhaps not as wary of such advancements as some scholars fear (Stolt et al., 2002; Campbell & Ross, 2003; Lernmark et al., 2004; Feuchtbaum et al., 2006; Feuchtbaum et al., 2007; Skinner et al., 2011). Still, trust and transparency in the health care system are both key principles of public health ethics and necessary for sustained

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67 Arguments for improved parent and HCP education and improved implied consent for NBS are drawn not only from my participants’ requests, but also from other empirical research examining parent, HCP, and lay public perspectives on NBS in Ontario (Hayeems et al., 2009; Miller et al., 2010b; Araia et al., 2012; Bombard et al., 2012; Hayeems et al., 2013).
success in public health programs (Thompson et al., 2006) and parent awareness, knowledge, and understanding is critical in fostering such trust.

Among the key informants who participated in my study and discussed issues around informed consent and express consent in particular, many articulated a host of concerns, which I interpreted as potential harms that could be introduced if an express consent process were implemented. Given the lengthy list of perceived harms, particularly regarding the need for improved HCP education and an effective consent implementation strategy, I further interpreted these perceived harms as contributing to the larger hesitation and ambivalence around pursuing an express consent approach for NBS in Ontario. Consequently, as the advisory committees explore the possibility of express consent – or another least restrictive approach – for NBS, their perceptions that the existing infrastructure is currently unable to support an effective approach to express consent that would support parental autonomy without sacrificing the public health goals of the program will arguably influence their thinking and final recommendations to the government. Given that express consent would be a new approach for the provincial NBS program, deliberations on such a shift in policy and practice should arguably incorporate empirical research from other jurisdictions, related Ontario-based research, and other theoretical, philosophical, and ethics scholarship that could provide insight into such a least restrictive alternative approach. In addition, public health ethics frameworks such as the one I present in this study could be useful in thinking through the known and unknown harms associated with an express consent approach to NBS. This public health ethics inquiry of key informant attitudes and perceptions of consent for NBS

68 And as I have noted repeatedly, a couple participants were clear that the province should not need to support such an approach given the public health nature of the NBS intervention.
generated a framing of consent that encourages considering a reconceptualization of express consent within a public health context to ensure that consent in practice is effective at meeting the needs of a socially, culturally, and economically diverse population. Moreover, the perceived strengths and limitations of express consent led to the interpretation of consent as a practice in need of a comprehensive evaluation prior to implementation.

6. Is the selection of individuals fair and appropriate?

As NBS is a province-wide program, all newborns are targeted and, therefore, all parents would need to give their express consent if such a policy were implemented in Ontario. The universality of the NBS program in Ontario is fair and appropriate. Therefore, by extension, that all parents would need to give express consent for their infant to be screened could be perceived as fair and appropriate as well, at least on the surface. However, to ensure a “[f]air distribution of burdens, risks and potential benefits” (Public Health Ontario, 2012: 4) of express consent, the express consent in practice arguably must be fair and appropriate as well to ensure that all parents are informed about NBS, understand the information provided, and understand the implications of their decisions. If, for example, express consent in practice privileges a certain demographic (e.g., highly educated, English/French speakers, Ontario/Canadian natives), however inadvertently, the argument could be made that the practice would no longer be fair and appropriate and would need to be modified accordingly. Consequently, the advisory committee should be cognizant of such possibilities during deliberations over approaches to informed consent for NBS.

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69 The original question read, “Is selection of participants fair and appropriate?” (Public Health Ontario, 2012).

Is it feasible?

Participants in my study questioned the feasibility of express consent for NBS due to logistical challenges perceived as temporarily insurmountable. That different jurisdictions around the world have different approaches to consent for NBS was perceived by some to be among the obstacles of an express consent process. Specifically, given that NBS awareness and education differ across jurisdictions, depending on how Ontario would implement an express consent process (e.g., prenatally or postnatally) participants worried that tourists or new arrivals to the city could be adversely affected by a consent process that does not consider such potential eventualities. Connected to this issue was the general tracking challenge of express consent: how to manage signatures (if there are signatures) and how to coordinate consent with Newborn Screening Ontario’s screening laboratory. Although the issue of tracking is a feasibility issue it can also be characterized as a potential harm, for any errors or mistakes in tracking could lead to an unscreened baby who might otherwise have been screened.

The storage of and future uses for dried newborn blood spots was considered by some participants who discussed these issues an added challenge to the feasibility of express consent, particularly regarding the possibility for future research initiatives involving the blood spots (see Chapter 7). When to introduce consent for storage and future uses to parents – and what that consent should look like, especially for the research dimension (e.g., blanket consent for any and all studies in the future or more circumscribed in its scope) – was

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70 The response to this question focuses on express consent.
identified not only as a potential harm to screening uptake, but also as a feasibility obstacle. Introducing additional steps for technicians in the laboratory was perceived by some as introducing an increased possibility for error.\textsuperscript{71}

Public health ethics analyses of public health initiatives require an anticipation of potential harms so that they may be preempted and/or mitigated (Tannahill, 2008; Public Health Ontario, 2012; ten Have \textit{et al.}, 2013). A least restrictive alternative such as express consent would arguably require similar consideration and attention prior to implementing such an approach to NBS. Participants who discussed tracking concerns conveyed a sense that they envisioned express consent as written and therefore introducing all the complications associated with such an approach (see Chapter 7). In our digital age, turning to technology to facilitate a consent process could potentially mitigate some of these concerns—although I recognize that technology is not without its own challenges that would need to be anticipated and addressed. A number of studies have documented efforts to obtain informed consent through online mechanisms (Flory & Emanuel, 2004). An online approach to express consent for NBS could be considered among the possible approaches to express consent — such as decision-aids, check boxes, and simple consent — for NBS. For example, an online consent database that includes the consent form in many different languages where a parent or parents can read the information and consent or dissent online at home or with their doctor. For those who dissent, a flag can be raised that results in a follow-up call to make sure parents understand what that decision means for their infant. Then, upon delivery the HCP responsible for conducting the heel prick can access the database and if the parents

\textsuperscript{71} How to overcome issues of feasibility with respect to express consent for NBS in Ontario for the screen and/or storage of and future uses for dried blood spots is outside the scope of this study.
have consented then they can proceed with the heel prick; if they have neither consented nor
dissented, consent can be obtained at the bedside prior to the heel prick and then entered into
the database. In keeping with the goal of this public health ethics analysis, express consent in
practice, whether technological or otherwise, would still need to achieve its theoretical goals
and should arguably be subjected to pilot testing in advance of implementation.

*Is it appropriate/warranted?*

This sub-question, within the context of considerations of informed consent for NBS
and express consent in particular, is particularly useful to consider within a public health
ethics framework given the overarching goal of finding a balance between public health
goals and individual rights and freedoms (see Chapter 2). A question that is arguably
worthwhile asking, in light of existing empirical scholarship on parent perceptions of and
attitudes towards express consent and implied consent, is whether express consent
mechanisms that mirror clinical and research ethics consent processes are appropriate for a
public health intervention such as NBS. Some studies have found that parents want to be
fully informed about all dimensions of NBS, regardless of whether the approach to screening
is mandatory or voluntary (Campbell & Ross, 2003). Some studies found, however, that
parents were ambivalent about express consent for the heel prick, worried about, for
example, the possibility of a cumbersome consent process and a lack of the appropriate
knowledge base to appraise the information to make an informed decision in the best interest
of their infant (Campbell & Ross, 2003; Detmar *et al.*, 2007; Moody & Choudhry, 2011).

Similarly, participants in my study were equally adamant that parents should be fully
aware of all dimensions of NBS independent from the approach to consent the government
decides to pursue for the screening program. Of the participants in my study who discussed storage of and research on dried newborn blood spots, many conveyed that while they have reservations about express consent as an approach to the newborn screen itself, most felt that implied consent is an inappropriate approach for storage and research uses that fall outside the scope of immediate infant health and program quality assurance purposes—a position arguably supported through other Ontario-based research exploring, in part, the public’s view on implied consent for storage and research of dried blood spots (Bombard et al., 2012).

Breaking down the NBS process into its component parts and seeking consent for storage and research separately held an initial appeal among some participants. Such tiered approaches to consent have been put forth as possibilities in the literature (Ross, 2010) and may be a worthwhile approach to discuss and assess within the context of consent for NBS in Ontario as well. Consideration should be given to each tier in terms of its ability to find the balance between achieving public health goals and objectives while also respecting parental autonomy. Among the main questions generated through my study (as I have previously articulated) is whether parents, if presented with consent for storage and future uses, will, as a result, decline screening as well. This empirical question is beyond the scope of this dissertation but the answer to this question will likely have implications for approaches to informed consent for NBS in Ontario in the future.72

As the province assesses its approach to informed consent for NBS, considering the options at hand within a public health ethics framework that takes parent preferences and perspectives into consideration in terms of the kind of approach to informed consent that they

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72 As new research findings emerge, the least restrictive approaches to NBS should be reevaluated and, if necessary, adjusted.
would prefer to have for NBS could be a helpful step in respecting and supporting parental autonomy within the broader public health mandate of the program. Given general parent support for expanded NBS, there might be an approach to consent for NBS that can respect parental autonomy yet also minimize the host of perceived consent-related harms identified by participants committed to achieving the broader public health goals and objectives of the program. With respect to the issue of storage and research for NBS, the NBS advisory subcommittee might find it helpful to apply the question of consent for research on stored blood spots to Public Health Ontario’s (2012) original analytic framework (see Table 1, column 4), as it was designed specifically for public health research initiatives (see Chapter 2). Such an exercise could be helpful during deliberations concerning how to proceed with informed consent moving forward.

Is it sufficient?

A number of the participants who spoke about informed consent did so within the broader context of improved NBS awareness, knowledge, and understanding among the general public and HCPs. While parent education is a critical component of informed consent, I interpreted my participants’ requests for improved community awareness of NBS as an important foundation upon which to introduce express consent. A number of participants spoke directly about their desires to work towards NBS being “commonplace” so that parents have heard of NBS, they know what it is, they know why it is conducted, and they know why blood spots are stored and used for de-identified research purposes. I would argue that establishing this basic level of knowledge and understanding in advance of parents facing an express consent decision would serve to support a more effective express consent
process not only in terms of supporting self-determination and autonomy, but also with respect to attaining the broader public health goals as well. Participants were clear that Newborn Screening Ontario has made, and continues to make, significant efforts to educate parents and HCPs about NBS. However, some participants revealed that they were discouraged by what they perceived to be a general lack of knowledge and understanding about NBS not only by parents but also HCPs. However, it is important to note that until the expansion of Ontario’s NBS program in 2006, NBS in Ontario had been conducted the same way for almost half a century. Theories of change process indicate that system change can take decades to become fully implemented (Fullan, 1985; Fullan, 2008). Consequently, education campaigns arguably need to be sustained and, according to my participants, multifaceted—not relying purely on written approaches to information dissemination but drawing on other visual forms of dissemination as well in order to support and facilitate system change.

8. Is community engagement warranted? Is it feasible? What level of engagement is appropriate?

Community engagement research on the topic of parental consent for NBS has already occurred in Canada (Bombard et al., 2012). This study, presented by Bombard et al. (2012), held 8 focus groups (n=60) in Toronto, Ontario and Montreal, Quebec. The objective of this community engagement initiative was to examine Canadian values with respect to parental consent for NBS, with a particular focus on the storage of and secondary uses for newborn blood spots (Bombard et al., 2012: 240). Findings from this study revealed participants’ support for the storage of blood spots for the purposes of confirmatory diagnosis, quality control, and anonymous research (Bombard et al., 2012). However,
participant views varied as to whether parental consent should be sought for future anonymous research (Bombard et al., 2012).

Another finding presented in this article focused on some study participants’ perception that it was the parent’s responsibility to self-educate about the various components of NBS (Bombard et al., 2012). More specifically, some participants felt that an information pamphlet with a few lines about the storage of and future uses for blood spots such as anonymous research with a link to the Newborn Screening Ontario website would be “sufficient” (p.243). After that, some participants felt it was the parents’ responsibility to self-inform about NBS (Bombard et al., 2012: 243). At this juncture it is important to note that Bombard et al. (2012) acknowledged that their community engagement study was comprised of “a highly educated and unrepresentative sample”73 and, as a result, recommended future community engagement research involving “underrepresented communities” (p. 245). This finding, with an emphasis on the dissemination of NBS information through written media and the perception that parents should self-inform about the multi-dimensionality of NBS contrasts directly with what some of my participants’ identified as potential obstacles in achieving an informed community. Some participants in my study were clear that purely written forms of communication such as pamphlets are insufficient when it comes to educating a diverse public about the various goals and objectives of NBS. Other participants felt that such a purely written approach to parent education would privilege certain members of a community over others, particularly the educated and otherwise socially advantaged parent.

73 With respect to education, 52 participants were characterized as having “some college or university and above”, while 8 participants had “high school and below” (Bombard et al., 2012: 241). In addition, participation in this study required that participants be able to consent in English (Bombard et al., 2012: 240).
As Newborn Screening Ontario, government advisory committees, and other researchers continue to explore the possibility of consent for NBS, community engagement initiatives could be a useful approach to address some of the perceived strengths and limitations of express consent for NBS provided participants from communities typically underrepresented in such research initiatives are involved. Existing scholarship on NBS and consent focuses on challenges of knowledge and understanding in conjunction with reading difficulty of the information materials and preferred approaches to information delivery among the highly educated (e.g. pamphlets and self-informing). Therefore, I would argue that future community engagement initiatives should consist of parents and expectant parents from diverse sociodemographic backgrounds representing a range of ethnicities, education and literacy levels, economic status, religious affiliations, and geographic locations (e.g., rural and urban representation) to explore the potential influence of social determinants of health and other sociocultural influences on an express consent process for NBS. In addition, individuals whose families have a history of some of the conditions on the screening panel should also be represented in such research initiatives.

Given the recurring themes in my study around issues of social justice in relation to informed consent, specifically the perception that consent may privilege certain members of society more than others, robust representation from individuals typically considered more difficult to recruit for such studies is imperative. Otherwise the study outcomes arguably risk reinforcing disparities that may emerge as a result of consent processes rather than work to mitigate them. If such diversity is not feasible, while the community engagement exercise may be fruitful in some important and illuminating ways, it arguably must be weighed
accordingly in light of other existing information and data, particularly as it pertains to health equity.

In addition, conducting community engagement research that includes the HCPs that would likely be involved in the consent process would also be important given that HCPs are considered critical to the practice of express consent. Incorporating HCPs in the process of designing and refining an express consent process will hopefully help foster HCP support for NBS and potentially increase the likelihood of success once consent is implemented. For if HCPs are given an opportunity to contribute to the process in terms of their perspectives and needs within the context of an express consent process for a public health initiative such as NBS, their involvement from the outset may serve to garner buy-in, minimize concerns that HCPs will pose a threat to “true” consent, and perhaps improve the likelihood that the approach to consent ultimately adopted by the province will be closer to achieving the sought after balance between realizing public health goals and objectives while also facilitating parental autonomy.

Drawing on themes of perceived harm connected to express consent that emerged in my data, pursuing community engagement strategies that try and assess how best people in the community like to receive and absorb information; how much detail they want to receive about NBS; when do they want to make decisions; and/or to evaluate a pilot project of a desired approach to consent could be invaluable in the process of identifying an effective and palatable least restrictive approach to informed consent for NBS for all key players involved, particularly parents. To achieve these ends, future study participants would arguably need to be educated not only about NBS, but also different ways of exercising their autonomy within
the larger public health context. Such a process could be invaluable in terms of evaluating different types of approaches to express consent by examining the type of information shared, whether the information is presented in a manner in which everyone can understand, the extent to which comprehension is retained, as well as different approaches to delivering that information. For instance, is written consent the most effective in terms of conveying the information? Is verbal communication preferred? What about computer-based consent programs? A combination of these options?

Although the majority of participants in my study did not reference community engagement with respect to the actual practice of informed consent, this framework is meant to guide governing bodies such as the advisory committees in terms of whether to implement express consent for various dimensions of NBS. This public health ethics framework question about community engagement suggests that a study could be conducted to evaluate the practice of a particular express consent approach for NBS prior to implementing it on a provincial scale. Given concerns regarding the effectiveness of express consent and its ability to achieve its theoretical goals in practice, community engagement research involving the intended consent intervention could be invaluable to ensure – prior to province-wide implementation – that the practice succeeds in achieving both autonomous decision making and population health objectives.

9. **What are the social justice implications of this initiative?**

A number of participants in my study appealed to the social determinants of health when reflecting on the potential harm that informed consent for NBS – both implied and express – could cause for certain cohorts of society. Some participants identified education,
literacy, income, poverty, and occupational class as among the social determinants of health perceived as contributing factors in a consent process and underscored the importance of considering socioeconomic position (income, education, and occupational class) when considering the benefits and risks of an express consent process.\textsuperscript{74} Participants stressed the importance of considering socioeconomic position in terms of access to health care and the ability to comprehend the information that would be conveyed within an express consent approach. In contemplating the prospect of express consent for NBS, many participants noted that such a process would likely have to occur during prenatal care. Some participants expressed concern that women in a lower socioeconomic position may be less inclined to seek prenatal care, and, therefore, they might miss the opportunity to give consent for NBS. Therefore, as the advisory committee considers the creation of a new consent process for NBS (or the improvement of the existing implied consent approach), it should consider the health care seeking behaviour of all women and incorporate a process that has the broadest reach. Otherwise, unscreened children within the context of a poorly implemented consent process could be more indicative of circumstance rather than parental autonomy.

In terms of information comprehension, some participants articulated concern that individuals with lower education and reduced literacy might have difficulty reading an express consent document (if such a process were in written form) and/or comprehending the information (whether oral or written approach to express consent). These participants felt that such lack of comprehension could result in some parents making uninformed decisions which, in addition to undermining parental autonomy, could potentially compromise infant

\textsuperscript{74} Some research that has been conducted in the area of research ethics also underscores the challenges of obtaining express consent among individuals with mental health challenges, such as depression and schizophrenia (Jimison et al., 1998).
health. Existing research that focuses on improving the process of express consent in clinical and research settings has found that gaps in knowledge and understanding following a consent process are not limited to individuals with lesser education, but rather that the knowledge gaps among these demographics are a little larger (Tait et al., 2005; Crepeau et al., 2011).

The social justice implications of consent can be perceived from an alternative perspective as well (though not a perspective introduced by my participants). If an express consent approach is not introduced for NBS on the basis that certain individuals or community groups would be marginalized, for example, one might argue that such groups, as a demographic, are being exploited as a reason not to implement express consent. Decision-makers and advisors could be perceived by some as using the fact that certain individuals in the community are marginalized to strip them further of their individual rights and freedoms; specifically, their right to make decisions regarding their healthcare and the healthcare of their children. One can argue that failing to honour the principle of respect for persons in a way that fosters autonomy perpetuates the marginalization experienced by certain members of a given community or population and in so doing can compromise their welfare. Perhaps social determinants of health should not preclude introducing express consent for NBS, but rather serve to highlight areas that would need special attention: efforts would need to be made to ensure that the mechanism of consent appealed to a very broad audience.

Participants in my study discussed their concerns about what they perceived to be potential logistical challenges introduced by an express consent process for NBS as well as concerns regarding the varying levels of HCP education regarding NBS, both of which I
interpreted as having implications for issues of social justice as well. More specifically, health care institutions vary in terms of their size, resources, staff, and location. The extent to which one can exercise one’s autonomy through truly informed decision-making is, I would argue, largely dependent on each HCP’s commitment to and investment in the consent process (e.g., some HCPs are more knowledgeable, more attentive, more respectful of autonomy, etc.) and the infrastructure of the hospital more generally. While all HCPs have the same ethical obligations with respect to consent, some may execute the process more effectively than others. As some participants stated, some hospitals in a given geographic area are more committed to NBS than others and some HCPs are more invested in the developments of NBS than others. Inevitably such institutional disparities will have implications for whether and how individuals are supported in exercising their autonomy within the context of informed consent for NBS.

Finally, I draw on Powers and Faden (2006) to underscore the arguably increased importance of attending to issues of social justice in instances involving parents and their children. Powers and Faden (2006) underscore their belief that children have a privileged position in social justice and that childhood health is critically formative and ultimately indicative of their future well-being. These scholars also recognize that privileging childhood health within a social justice framework necessarily demands that parent health be given a priority as well:

The focus in social justice on securing the well-being of children also requires placing a priority in public health on the health of parents … That the health of children and their subsequent well-being turns critically on the well-being of women during pregnancy and childbirth is only the most obvious way in which the health of parents are important to the health of their children. (Powers & Faden 2006: 94)
While children’s health is privileged, there is an acknowledgment that children’s health is dependent on parental health: as an initial point of departure children and parents are presented as a unit rather than juxtaposed from the outset as adversaries. Therefore, I agree with Powers and Faden (2006) that promoting childhood health requires attending to parental well-being as well. In addition to addressing the physical health needs of parents, efforts should also be paid towards cultivating, through an express consent process, the dimensions of well-being such as respect, reasoning, and self-determination necessary to enable parents to make informed healthcare decisions for their children that not only promote their child’s immediate health, but will also serve to preserve the possibility for the future growth and development of well-being in their children (Powers & Faden, 2006).

The principle of social justice considered through a public health ethics lens asks us to contemplate what a least restrictive approach to express consent would need to look like to promote the collective well-being of parents and their children. Within the specific context of informed consent for NBS, what would the practice of consent need to look like to facilitate reasoning and self-determination in all parents with diverse socioeconomic and sociocultural backgrounds, which could then affect infant and childhood health? The public health ethics principle of social justice requires that we ask these questions of the practice of express consent in deliberations about consent policy and practice for NBS. It is essential to think through what injustices are possibly produced through express consent, implied consent, or other consent processes that fail to meet their theoretical ideals in practice. Such injustices, conceived within the comprehensive interpretation of well-being put forth by Powers and Faden (2006), can pertain to health, reasoning, respect and self-determination.
10. What are the potential long-term consequences?

If an express consent process is implemented for NBS without being properly vetted for functionality within a public health context, the potential long-term consequences can be grave, particularly if consent confusion and/or malfunction leads to an increased number of infants being diagnosed symptomatically rather than through the newborn screen. However, preventing a population from moving towards a greater understanding of NBS through a supported express consent process out of the fear that parents will decline screening if presented with such a consent – a fear that is currently unsubstantiated in Ontario – is arguably indefensible. Still, if the advisory committee were to conclude, after a comprehensive public health ethics analysis of the benefits and risks of consent for NBS, that express consent is arguably not appropriate at this time given that the infrastructure necessary to support such an intervention does not currently exist or is not sufficiently robust, then one potential long-term consequence of rendering such a conclusion is that the decision could translate into inaction rather than setting express consent as an achievable goal for the near future. In the meantime, as a more comprehensive least restrictive approach to consent for NBS is being designed to account for the multidimensionality of expanded NBS (or as improvements are made to the current implied consent approach), more direct communication about the storage and research dimensions of NBS in the printed informational materials given – at least in theory – to parents is imperative to begin the process of improving the effectiveness of implied consent as an approach to fostering parental autonomy.

As explicated in this public health ethics analysis, express consent is not the only way to promote autonomous decision-making. Autonomy in public health contexts can be
fostered and promoted through other least restrictive alternatives (Nuffield Council on Bioethics, 2007). Opt-out programs, implied consent, and community consensus are also put forth in the public health ethics literature as legitimate least restrictive alternatives (Upshur, 2002; Public Health Ontario, 2012). However, a crucial component necessary to respect parental autonomy through such least restrictive alternatives as opt-out or implied consent mechanisms is a public awareness and recognition of the fact that opting-out is an option. Public health ethics analyses are iterative processes and as new data becomes available and as efforts are made to improve public education, least restrictive approaches to consent for NBS that can succeed in maximizing benefits and minimizing harms for both public health and parental autonomy should be explored and reevaluated regularly.

Chapter Conclusion

One of the goals of this analysis and interpretation was to contribute to the ongoing discussion in Ontario about informed consent for NBS. My descriptive, analytic, and interpretive themes work collectively to challenge the often seemingly taken-for-granted assumptions embedded in the notion of express consent within a public health context such as NBS. A common refrain familiar within much of the public health ethics literature currently in circulation is,

If data show that a voluntary screening program will test essentially the same number of individuals as a mandatory one, because almost no one refuses testing when asked, then it would be ethically improper to implement a mandatory program. (Kass, 2001: 1780)

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75 I have also drafted a preliminary figure that endeavours to capture the spectrum of autonomy and state infringement within the context of medical public health programs (see Appendix E).
However, such a comment assumes that the voluntary nature of an intervention such as consent actually accomplishes its objectives. Framing the policy and practice of consent as a least restrictive alternative and highlighting its benefits and harms underscores that consent is arguably not something that can simply be tacked on to a public health program, but rather needs to be examined and evaluated as would any other public health initiative.

In Ontario, implied consent is, according to the *Health Care Consent Act* (1996), an approach to informed consent and therefore the same standards must be met in terms of knowledge, understanding, and non-coercive participation. My research (see Chapter 6), coupled with the study findings of Bombard *et al.* (2012) and Araia *et al.* (2012) suggest that there is work to be done in Ontario before Newborn Screening Ontario can feel confident that an absence of parental refusals can be interpreted confidently as consent. Informed consent for a public health program (whether implied or express), just like pharmaceuticals, biotechnologies, and counselling programs for example, should arguably not be introduced on a massive public health scale without taking steps to identify and address possible harms.

In jurisdictions that have already implemented express consent, if the process does not achieve the goals and objectives of consent, then one could question whether express consent should have been implemented in the first place. Public Health Ontario’s (2012) public health ethics framework provides a series of questions that can aid in a comprehensive consideration of various least restrictive approaches to NBS including implied consent. The application of my study findings to a public health ethics tool aimed to guide professionals engaged in public health initiatives think through least restrictive alternatives from a public health ethics lens arguably serves to bridge the gap between the ever-present tensions in
public health initiatives between individuals and communities. Current debates around the topic of express consent for NBS focus almost exclusively either on liberal or consequentialist arguments for or against express consent for NBS. Framing express consent as a least restrictive alternative that can be analyzed using a comprehensive public health ethics frameworks arguably shifts the conversation towards a third lens of analysis that aims to bridge the impasse through the design and implementation of an approach to informed consent for NBS that succeeds in achieving the public health goals of the program while also respecting parental autonomy.
Chapter 9. Conclusion

Chapter Overview

This qualitative case study explores, analyzes, and interprets participant attitudes and perceptions of informed consent, both implied and express, for NBS in Ontario through a public health ethics lens. A consistent theme in my data was a perceived disconnect between the ethical concept of informed consent and perceptions of how it actually unfolds in practice. Participants’ attitudes towards implied and express consent for NBS challenged conceptions of consent as a presumed good that promotes parental autonomy. Rather, some participants articulated their concern that the theoretical ideals embodied in the principle of consent do not necessarily translate in practice. Within the context of express consent in particular, this perceived disconnect between theory and practice introduced what I interpreted as a series of perceived harms that threaten to put infant, parent, and societal health at risk.

At a thematic level I characterized these harms according to consent process, content, delivery of information, logistical obstacles, and the illusion of choice. Applying a public health ethics lens to this analysis facilitated a framing of consent through four public health ethics principles: least restrictive alternatives, effectiveness, autonomy, and social justice. When further interpreted within the broader public health ethics challenge of promoting population health while also respecting individual rights and freedoms, I interpreted my participants’ perceptions of consent as harm-causing, not only in terms of compromising the
goals and objectives of the public health program, but also potentially undermining parental autonomy as well.

In this final chapter I synthesize my contribution to the public health ethics literature on the topic of informed consent for NBS. Based on my findings, I also make a few modest recommendations for the current Maternal-Child Screening Committee and Newborn-Child Screening Subcommittee as well as some ideas for future research. I then outline the limitations and strengths of this study followed by a brief conclusion.

**Study Contributions**

This public health ethics inquiry regarding consent for NBS in Ontario revealed that key informant fears regarding informed consent, and express consent in particular, extend far beyond parental refusals. A more robust conceptualization of the perceived harms of express consent for NBS is arguably useful in two ways. First, as a program that is striving to gain the public’s trust and participation, presenting a more comprehensive, complex, and nuanced argument regarding the perceived challenges of express consent for NBS will likely be more palatable to parents than currently existing arguments against express consent such as those proposed by Feuchtbauern et al. (2007) that insist parents cannot be trusted to make healthcare decisions in the best interest of their infants. Replacing such an arguably aggressive point of departure with an articulation of the multidimensionality of the concerns connected to consent for NBS not only for infant and population health, but also parental autonomy may resonate more positively with parents if they are not made to feel “unfit” to make healthcare decisions on behalf of their children. Moreover, many participants in my study insisted that parents need to be better informed about all dimensions of NBS in Ontario. This belief,
juxtaposed against concerns that if parents are faced with an explicit decision within the context of express consent that they might refuse screening on behalf of their infant, suggests that the main issue in need of a resolution is skepticism around express consent rather than an unwillingness to inform parents about NBS and/or to honour and respect their autonomy.

Establishing consent as a least restrictive approach to NBS arguably creates a structured frame within which to address this skepticism through a comprehensive public health ethics analysis. Examining consent through a public health ethics lens that strives to ensure that public health programs, interventions, policies, and practices are ethically sound in their ability to balance public health goals against individual rights and freedoms is an approach that seeks to ensure, to the extent possible, that the theoretical ideals of consent are met in practice. Participants in my study articulated their doubts that implied consent, as currently practiced in Ontario for NBS, succeeds in achieving the required standard of informed consent. They were also equally wary of an express consent approach for NBS, concerned that it would compromise the screening program and its population health goals, the principle of autonomy, or both. Given the public health nature of NBS, informed consent within this context is, I would argue, a public health practice. Therefore, considering, assessing, examining, and evaluating informed consent through a public health ethics lens is useful because it becomes an intervention in and of itself that must meet certain requirements rather than simply a requirement that must be considered and/or met. Moreover, whether the ultimate approach to informed consent for NBS in Ontario is implied or express – or some other possible hybrid between the two that seeks to achieve the aforementioned balance
between uptake and autonomy – the practice should be truly a least restrictive alternative rather than the mere illusion of one.

In addition, my public health ethics analysis and interpretation of consent for NBS corroborates, reconfirms, and builds empirically on existing framings of NBS programs as wrestling with the tensions between protecting and promoting parental autonomy while also achieving the population health goals of the program (Hargreaves et al., 2005b; Newson 2006). My public health ethics analysis and interpretation of consent for NBS also offers an alternative approach to thinking about consent for NBS that extends beyond more traditional characterizations of the public health ethics dilemma as either meeting the goals and objectives of the public health program or respecting individual rights and freedoms as preserved through self-determination and autonomy. Furthermore, framing consent as a least restrictive alternative for a medical public health initiative with embedded harms of its own may allow for future inquiry into the reconceptualization of an express consent approach for a public health program such as NBS. Scholars have documented shortcomings of express consent in clinical and research contexts and, increasingly, in public health contexts as well (Faden et al., 1982; Holtzman et al., 1983; O’Neill, 2004b; Hargreaves et al., 2005a; Hargreaves et al., 2005b; Dawson, 2005; Nijsingh, 2007; Nicholls, 2010). One might argue that shortcomings of a consent practice in the context of a public health program can have more far-reaching implications not only for infants, but also parents and society more broadly. Therefore, working towards consent as an effective least restrictive approach for NBS, whether implied or express consent, will be important.
Finally, a public health ethics analysis of implied and express consent as least restrictive approaches for NBS is a modest first step towards answering Faden and Shebaya’s (2010) call for an examination of potential embedded harms associated with least restrictive alternatives in public health. Moreover, considering the importance of such public health ethics principles as least restrictive alternatives, effectiveness, autonomy, and social justice within the context of informed consent will hopefully be useful in continued efforts to improve the effectiveness of such least restrictive alternatives. While this inquiry focuses on participant perceptions around the policy and practice of informed consent for NBS in Ontario, the challenges identified by participants in my study are arguably not unique to the province. A public health ethics approach to consent may be useful for other jurisdictions either reconsidering or striving to improve their respective NBS consent policies and practices.

**Recommendations Moving Forward**

Based on the findings from my study, I offer a few modest recommendations to the current Maternal-Child Screening Committee and Newborn-Child Screening Subcommittee. The mandates of these provincial advisory committees include addressing ongoing and emergent issues pertaining to consent for NBS, the storage of newborn blood spots, and the future use of blood spots for research (Born Ontario, 2013a). I have also outlined a number of possible next steps in terms of research initiatives that could build on my study findings and offer additional insights into the longstanding question of informed consent for NBS.
Recommendations to the Advisory Committees

This study revealed various and conflicting understandings of implied consent within the context of NBS in Ontario. Although some participants in my study maintained that the current practice of implied consent for NBS in Ontario appeared to serve as a mandatory screening equivalent, others aligned implied consent more closely with an approach to informed consent. The language of consent used in the NBS literature is similarly varied (Hargreaves et al., 2005b). Given the diversity reflected in my participants’ perceptions of what constitutes implied and express consent, even among members of the advisory committees, I recommend that the advisory committees strive to establish (or confirm and disseminate) a shared, consistent language around consent approaches for NBS including labels, definitions, and goals and objectives of each approach. Such efforts would likely be beneficial for future consent debates and deliberations. Similarly, given that I identified a lack of consensus among my participants in terms of what implied consent means, how it should be implemented, and whether it is an appropriate approach for NBS in Ontario – coupled with similar disagreements around what would constitute express consent for this public health program – I recommend that the advisory committees consider inviting legal scholars who specialize in ethics, public health ethics, and/or research ethics to become permanent members of the committee. Such expertise could provide an invaluable contribution to current and future debates and policy deliberations around informed consent for expanded NBS in the province.

Even though participants recognized and acknowledged Newborn Screening Ontario’s efforts to improve NBS education in the province, many still expressed the need for
improved NBS education for parents and HCPs. Specifically, some participants expressed feeling discouraged that, from their perspectives, many HCPs have seemingly not yet embraced fully the changes to the province’s NBS protocol and, as a result, many parents remain insufficiently informed about NBS. Institutional shifts in practice can take decades to become fully implemented and succeed in transforming practice and achieve systemic change (Fullan, 1985; Fullan, 2008). Since NBS in Ontario had been conducted the same way for 40 years until the program expanded in 2006, perhaps it is not surprising that all HCPs have not yet fully adjusted to the changes. To facilitate the institutionalization of expanded NBS in Ontario, the advisory committees might consider enlisting the help and input of change process theory experts to support HCPs and their institutions with the ongoing program change. While it might take some time for the expanded program to become commonplace among HCPs, professionals trained in such institutional changes could conceivably offer insights to expedite the process.

Given the perceived lack of parent awareness, knowledge, and/or understanding of NBS, the advisory committees might consider suggesting a multimedia approach to information dissemination to extend the reach of NBS education across the province. Also, the printed informational materials for parents (e.g., NBS brochures and parent information sheets (see Appendices F-G)) in circulation at the time I conducted my study make no mention of the technology’s ability to detect carriers of sickle cell anemia or existing processes for long-term storage of and future uses for dried newborn blood spots. Given that most of my participants were emphatic that parents need to be fully informed about all dimensions of Ontario’s NBS program, the advisory committees might consider
recommending that a more transparent and comprehensive overview of Ontario’s expanded NBS program be communicated to parents via these printed “take-home” materials. Both initiatives may prove useful in cultivating a more informed population. Similarly, Newborn Screening Ontario’s website is comprehensive in terms of the information that it provides. However, the “Overview” drop-down option in the parents’ section of the website does not include the program’s ability to identify carrier status for sickle cell anaemia, nor does it outline its practices regarding the storage of and future uses for dried newborn blood spots (Newborn Screening Ontario, n.d.c). While this information can be found elsewhere on the website, and access to this information is facilitated through embedded hyperlinks in the Overview section, I would argue that a succinct overview of all NBS practices accompanied by clear, simple steps parents can take if they wish to opt-out of certain dimensions (e.g., long-term storage) or opt-in to certain dimensions (e.g., learning their infant’s carrier status) should be clearly outlined in the Overview for easy, immediate, and succinct access.

Recruiting healthcare education experts with specific expertise in devising education tools to disseminate complex information to an economically, socially, culturally, and cognitively diverse population could be invaluable for future efforts to improve parent awareness, knowledge, and understanding of NBS. Given the study finding that informed consent – both implied and express – can have social justice implications and be more or less effective depending on its design and implementation, a healthcare education expert may be a useful asset to aid in crafting an approach to informed consent that would facilitate self-determination across a population.
Participants in my study articulated possible harms related to logistical and feasibility challenges of a province-wide approach to express consent. Software design and information technology specialists, in conjunction with healthcare education scholars, the NBS advisory committee, and the Newborn Screening Ontario team, could help envision effective approaches to the design and implementation of informed consent that could succeed in preempting harms related not only to issues of logistics and feasibility, but also knowledge and understanding. Incorporating such experts in the development of education and/or consent initiatives from the outset could provide meaningful contributions to the design and dissemination of NBS information within education and consent contexts.

Finally, given that I framed approaches to informed consent within the context of public health programs as least restrictive alternatives and used Public Health Ontario’s (2012) public health ethics framework to offer a comprehensive exploration of the ethical justification of informed consent for NBS, I recommend that the advisory committees explore the strengths and weaknesses of using such a framework as a group. This framework offers the advisory committees a new lens through which to examine different least restrictive approaches for the many dimensions of NBS, including the storage of and future uses for dried newborn blood spots. Questions to ask include, What issues does this framework surface when implemented within a group context? Does the framework succeed in creating an opportunity to resolve tensions around the issues of consent? Does the framework succeed in facilitating a negotiation of an approach to informed consent that could both foster informed, autonomous decision-making and maintain the goals and objectives of the NBS program? Understanding whether and how Public Health Ontario’s
(2012) framework succeeds as a group tool for discussions and deliberations for consent policy and practice will inform whether it succeeds in providing an alternative framework for considering the ethical justification of consent for a public health initiative.

**Recommendations for Future Research**

There are a number of possible next steps in terms of future research initiatives that could build on the study findings and insights generated from my inquiry. A natural first step might be to design a study that draws on my public health ethics framing of informed consent as a least restrictive alternative for a public health intervention such as NBS to ascertain the perspectives and attitudes of a representative study sample of parents and/or expectant parents towards different least restrictive approaches for expanded NBS in Ontario, including but not limited to implied and express consent (see Appendix E). A study that incorporates such key public health ethics principles as least restrictive alternatives, effectiveness, autonomy, social justice, transparency, and trust within the context of informed consent for NBS could be instrumental in understanding how parents and expectant parents perceive different possible practices of consent for NBS and their preferences regarding the amount of information they wish to receive about the various dimensions of NBS; when and how (e.g., in what format) they wish to receive the information; and whether they identify any risks or potential harms (either for the program or for parents) associated with implied or express consent practices. The findings from such a public health ethics inquiry would be informative in their own right as Ontario continues to evaluate different approaches to consent for NBS. In addition, these findings would also be useful as a point of comparison to identify similarities and differences among the perspectives of lay parents and expectant
parents and those of the key informants who participated in my study, as the empirical literature has found that parents and health care providers often perceive ethical issues related to NBS differently (Rothwell et al., 2010; Tarini et al., 2008).

In considering additional future research directions that could extend from the findings generated through my public health ethics inquiry, I identified three overarching themes – generated from my analysis and interpretation – that arguably capture the current gaps in consent scholarship for NBS. First, does implied consent for NBS in Ontario meet the standard of informed consent and is it effective in fostering self-determination among parents? Second, would an express consent approach to NBS in Ontario succeed in fostering self-determination and informed decision-making without compromising the public health goals and objectives of the program? Third, would introducing express consent for storage of and future uses for dried newborn blood spots (e.g., research, forensics) compromise screening uptake within an implied or express consent approach to NBS? These empirical research questions are each significant in scope and the answers integral to the continued evaluation and evolution of an effective least restrictive alternative for NBS in Ontario that succeeds in achieving a balance between protecting individual rights and freedoms and achieving the public health goals and objectives of the program. I could not begin to offer an exhaustive list of the many different studies that could be undertaken for each of these questions. However, I will propose a few possible directions based on my study findings.
1. Does implied consent for newborn screening in Ontario meet the standard of informed consent and is it effective in fostering self-determination among parents?

The practice of implied consent in Ontario is one issue that generated much debate among participants in my study. Points of contention included whether parents are sufficiently informed about NBS and whether parents possess the knowledge and understanding of NBS necessary to meet the standard of informed consent so that HCPs can confidently infer parental silence on the issue of expanded NBS as consent. An ethnographic study that combines prenatal and post-delivery observations of parental care meetings and heel pricks to see when, whether, and how NBS information is introduced to parents would provide invaluable insight into how implied consent is operationalized in Ontario.

Similarly, participants in my study challenged the effectiveness of implied consent as a least restrictive alternative that succeeds in generating informed, autonomous, parental decisions. A study designed specifically to assess parental awareness, knowledge, and comprehension of the many facets of NBS (e.g., storage, research) currently captured in the implied consent approach for NBS could be an invaluable empirical contribution to the ongoing debate of the effectiveness of implied consent for NBS as an approach to informed consent. In addition to assessing parental awareness, knowledge, and understanding, it would arguably be important to ask parents whether and how they “gave” their consent to have their infant screened.

A study that explores attitudes and perceptions of implied consent from the perspectives of HCPs charged with conducting the heel pricks would complement empirical studies involving parents: how do these HCPs define implied consent for NBS?; what do they tell parents about NBS, if anything?; and do they interpret parental silences on the issue of
NBS as informed consent? Such a study should recruit a representative sample of HCPs who would be charged with implementing the least restrictive alternative decided upon by the province to increase the likelihood of buy-in and a successful execution.

If studies find that implied consent as currently implemented does not succeed in fostering self-determination among parents for NBS, can the practice of implied consent be modified in an effort to respect parental autonomy while also maintaining high screening uptake?

2. Would an express consent approach to newborn screening in Ontario succeed in fostering self-determination and informed decision-making without compromising the public health goals and objectives of the program? What would express consent within a public health context need to look like to achieve such a balance?

A number of participants in my study articulated their concerns that while the ideals of express consent are laudable, they questioned the extent to which they are realized in practice and more importantly the impact that an ineffective approach to consent on a provincial scale could have on infant health specifically and population health more broadly. A pilot study designed to examine the effectiveness of express consent as a least restrictive alternative for NBS could both draw from and build on my applied, public health ethics analysis of express consent in an effort to identify an approach to express consent that succeeds in meeting the information and comprehension needs of parents without compromising the goals and objectives of the program.

Such a study might also include a more in-depth public health ethics exploration of the ethical justifications for a reconceptualization of what express consent looks like within the context of public health programs such as NBS. After identifying possible least
restrictive alternatives for NBS that hold promise for mitigating the ethical tensions inherent in public health, whether in the form of a decision aid or simple consent for example (see Chapter 7), and analyzing them through a public health ethics framework, such as the one provided by Public Health Ontario (2012), researchers could then conduct a pilot study to evaluate the effectiveness of these alternatives’ abilities to achieve the public health ethics balance of maintaining the goals and objectives of the NBS program while also supporting parental autonomy through informed decision making. Such a pilot project would also surface any unanticipated harms or other outcomes that may need to be addressed or considered in advance of widespread implementation.

3. Would introducing express consent for storage of and future uses for dried newborn blood spots (e.g., research, forensics) compromise screening uptake within implied or express consent approaches to newborn screening?

My research also introduced key informants’ fears around parents’ perceptions of informed consent for the storage of and future uses for dried newborn blood spots. Specifically, although participants who discussed these issues were adamant that express consent for the storage of and future uses for dried newborn blood spots was necessary, they questioned whether introducing effective informed consent for such purposes that extend beyond immediate infant health and program evaluation/quality assurance would have a negative effect on screening uptake and, as a result, compromise the health goals of the program (see Chapter 7). An empirical study designed to examine when and how parents would like to receive information about storage of and future uses for the dried newborn blood spots (including research) and whether such information would deter parents from having their infant screened altogether would be useful information to inform future
deliberations on informed consent for NBS. Currently in Ontario, the dimensions of storage of and future uses for dried newborn blood spots are captured through implied consent; however, this information is not found in any of the printed informational materials given to parents (see Appendices F-G). If implied consent within the context of a public health intervention is a least restrictive alternative that could foster self-determination through informed decision-making while maintaining high screening uptake, efforts could be made to assess empirically whether improved transparency of all dimensions of NBS in Ontario through various information outlets succeeds in improving the quality of implied consent without negatively affecting the primary newborn screening goals and objectives. Finally, a study that compares screening uptake outcomes as well as storage and future use participation obtained through implied consent processes and express consent process might generate interesting findings that could also shed light on the consent debate for NBS in Ontario.

Separate from these three overarching research questions, one dimension of consent that was relatively absent from both my interviews and the extant literature on NBS and consent is the approach to express parental consent for follow-up testing after infants have screened positive for one of the conditions on the panel. In light of recent findings suggesting that parents in Ontario are largely supportive of expanded NBS, even if no treatment exists (Hayeems et al., 2013), it would be interesting to research what choices parents are given at the time of follow-up testing and whether and how consent processes at the time of the heel prick could be considered in conjunction with consent processes at the time of follow-up.
Finally, in considering the sample demographics of future research initiatives involving parents, diverse and representative study samples are imperative. Researchers should take care to recruit members of the community typically underrepresented in research studies to ensure that the social justice implications of express consent can be appropriately considered so as not to perpetuate information and/or consent preferences of some members of society at the expense of others.

**Study Limitations and Strengths**

The goal of this exploratory, qualitative case study was to describe, analyze, and interpret participants’ perceptions of and attitudes towards informed consent for NBS in Ontario from the perspectives of individuals involved directly or indirectly in Ontario’s NBS program. The nature of exploratory case study research demands a certain level of description in order to establish a foundation upon which to generate new insights and interpretations (Merriam, 2009). The public health ethics lens that I used for this inquiry facilitated an interpretation of informed consent as a least restrictive alternative that could cause harm. The descriptive component of this dissertation was critical for three reasons. First, the descriptive dimension offers insight into how key informants perceive implied and express consent for NBS in Ontario with a particular focus on the vocabulary and phrasing used to convey their attitudes. (The words used by participants in my study to describe and discuss informed consent are crucial to the analysis and interpretation of the data.) Second, the descriptive component of my analysis underscores how the attitudes and perceptions of key informants in my study reflect the tensions inherent in public health and public health-inspired initiatives. Finally, the description showcases participants’ perceptions of the
benefits and harms of both implied and express consent articulated in a manner that resembles any other critical examination of a public health intervention that operates – or could operate – on a province-wide scale. Analyzing and interpreting participants’ narratives through a public health ethics lens allowed me to use the empirical data to reformulate conceptually the core concept of informed consent within a public health context such as newborn screening. My dissertation draws on this lens to offer one interpretation of my data. All qualitative research projects allow for multiple interpretations and, therefore, there are likely additional ways of interpreting my data using different methodological and theoretical lenses.

Issues of confidentiality, conflict of interest, and recall bias emerged as challenges in eliciting information from some participants who participated in my study. Participants who served as advisors on one of the NBS committees were bound by confidentiality agreements and, therefore, could not speak to me about the specifics regarding the debates and discussions around consent for NBS in Ontario. In addition, a number of participants were purportedly conducting their own research on NBS issues similar to those introduced in my study, which, as a result, may have affected the extent to which they shared with me their complete perspectives and attitudes towards ethical issues related to NBS in Ontario and informed consent in particular:

Your questions are very much overlapping with a lot of the current research that I’m involved in, so that’s what makes it tricky. So there are some things that I know that I haven’t said because I can’t reveal them, and there are some committees that you know, I have a privacy agreement with that I can’t talk about, so I mean that’s … I guess one of the challenges of your work is that it … um … you know … I know there’s things I haven’t told you but I can’t because of privacy so I have to be vague and … at some times. And it’s … as much as you’d like to be more honest … I don’t think … most of what I’ve told you I’ve been able to be pretty honest about, but the inner workings of a
committee you’re not allowed to really talk about those. (#3, advisory committee)

Also, a few participants who served on earlier iterations of the various government advisory committees had had a significant time gap between their committee work and/or the program’s expansion and their interview with me. As a result there may have been some issues of recall bias in terms of their personal views at the time, generalities shared about what was discussed in committee meetings, and/or specific events or people that contributed to the program’s expansion. That said, since this analysis and interpretation focuses specifically on participants’ own attitudes and perceptions of consent for NBS in Ontario, recall bias should not really be considered as a significant limitation to this inquiry.

Ultimately, the goal of my analysis was not to generalize participants’ perspectives on the issue of informed consent for NBS, but rather to draw on participants’ narratives to offer a reinterpretation of informed consent as an intervention that should meet the same standard of effectiveness as other public health initiatives. Public Health Ontario’s (2012) public health ethics framework provides a comprehensive analytic tool to consider the strengths and limitations as well as the benefits and burdens of informed consent processes for a public health program such as NBS.

No one study can examine every dimension of a given research topic. The exploratory nature of my inquiry coupled with my analytic and interpretive focus on a public health ethics reconceptualization of informed consent necessarily meant that other important ethical issues of NBS worthy of inquiry, such as the storage of and future uses for dried newborn blood spots, were not addressed as comprehensively as they would have been had they been the target focus of the inquiry from the outset. As articulated in the
“Recommendations for Future Research” section of this chapter, my analysis and interpretation has arguably introduced new findings and insights that can serve as the basis for additional future research on the topic of informed consent for NBS specifically and public health initiatives generally, as well as future analyses of other ethical issues directly related to NBS in Ontario (e.g., storage of and future uses for dried newborn blood spots).

Other strengths of this inquiry include the public health ethics lens of analysis that led to an interpretation of informed consent as a least restrictive alternative that could, like other public health initiatives, introduce (unforeseen) harms. In addition, a public health ethics analysis that draws on an applied public health ethics analytic tool, such as the one put forth by Public Health Ontario (2012), offers a new approach to debating the ethical justification of informed consent for NBS that breaks away from the two most common, yet polarizing, philosophies used to debate informed consent within the context of NBS: liberal and consequentialist. The framework I present in this dissertation for thinking about informed consent can accommodate diverse worldviews and offers the possibility of moving away from these two extremes with the ultimate goal of establishing a balance between enabling self-determination through informed decision making and achieving the public health goals of the program.

Concluding Remarks

In 2006 the Ontario government expanded the province’s NBS program at a time when academics were challenging the ethical justifications of mandatory screening programs as well as those that operate according to implied consent. Yet, Ontario continued with the implied consent approach that had been in place since the program’s inception in the 1960s.
Implied consent within the context of the expanded program includes the heel prick, newborn screen, storage, and de-identified research (Bombard et al., 2012; Newborn Screening Ontario, n.d.f). Now, more than six years later, addressing the ethical dimensions of consent for NBS is part of the current advisory committees’ mandates (Born Ontario, 2013a; 2013b). My research provides perspectives from four groups of individuals connected directly and indirectly to Newborn Screening Ontario: advisory committee members, HCPs at the treatment centres, advocates, and legal experts. Independent from my analysis and interpretation, at a very descriptive level, understanding perceptions of these groups of people will hopefully contribute to the discussion and debate around consent for NBS in Ontario.

As for the applied dimension of my study, the use of a modified version of Public Health Ontario’s (2012) public health ethics framework for my inquiry served two critical purposes: 1) to serve as an analytic tool to think about informed consent for NBS; and 2) to advance public health ethics scholarship by challenging the principle of least restrictive alternatives for public health programs, interventions, policies, and practices by analyzing an example of one such approach, namely express consent, according to the standards to which other public health initiatives are held.

By submitting the practice of express consent to this analytic public health ethics framework (Public Health Ontario, 2012), I provide insights into the prospect of consent for NBS that might otherwise have been missed had NBS been applied to this framework with the issue of consent addressed solely in one question. More specifically, the issue of consent in the original framework is limited to one question, #7: “Is individual informed consent
warranted? Is it feasible? Is it appropriate? Is it sufficient?” (Public Health Ontario, 2012). While these are important questions, they do not provide a sufficient framework to challenge the often taken-for-granted assumptions of the practice of consent itself. The questions do not provide an opportunity to think about, articulate and subsequently weigh the benefits and harms of the consent process itself. Applying the practice of express consent as a least restrictive alternative to the framework on the other hand, offers an opportunity for a more comprehensive and robust analysis of consent as a least restrictive approach for a public health program such as NBS.

Increasingly studies are showing that implied consent, at least as currently practiced in Ontario, is likely not supportable as an all-inclusive approach to NBS (Bombard et al., 2012). The challenge to implied consent in Ontario is arguably further supported by Araia et al.’s (2012) Ontario-based survey study, which found that out of 750 mothers who completed the survey, “only 35% responded correctly to a question designed to test whether they were aware that parents have a choice not to have their infant undergo NBS; many mothers responded ‘don’t know’ (46%)” (p.4). Other findings from Araia et al.’s study (2012) that question the effectiveness of Ontario’s implied consent process as an approach to informed consent include findings that show that only 35% of the mothers correctly answered a question that suggested they understood what a false positive screen result meant and less than 15% of those surveyed recalled learning about the storage of blood spots and the future uses of those blood spots. My findings, which offer the key informant perspective on issues relating to consent for NBS in Ontario further corroborate an argument for an adjustment of some kind to the current practice of consent for NBS. The question remains as to the best
approach to remedy this issue and whether informed consent is the answer.

By framing the practice of express consent through a public health ethics lens and applying it to a modified version of Public Health Ontario’s analytic public health ethics framework (2012), regardless of one’s theoretical or philosophical allegiances, the analytic lens turned on the practice of consent itself as a least restrictive alternative provides a useful analysis and generates important issues worthy of future consideration prior to implementing a province-wide consent process. In the spirit of public health ethics, which strives to reach a balance between libertarian and consequentialist philosophical poles, I would argue that challenging the practice of consent and highlighting potential areas of weakness that could cause harm to individuals and by extension communities demonstrates an effort to find a middle ground rather than falling strictly on one side of the debate or the other. This lens, while it surfaces potential shortcomings of consent, it brings them to the surface so that they can be mitigated prior to implementation. As advisory committees continue to discuss and deliberate approaches to consent for NBS, this analytic framework may be a useful tool to incorporate into the debate.
References


## Appendices

### Appendix A: Sample of Jurisdictions and their Respective Approaches to Consent for Newborn Screening

<table>
<thead>
<tr>
<th>Country or State/Province</th>
<th>Mandatory</th>
<th>Implied/Opt-Out</th>
<th>Explicit/Express</th>
<th>References</th>
</tr>
</thead>
<tbody>
<tr>
<td>New Zealand</td>
<td></td>
<td>X</td>
<td></td>
<td>Kerruish <em>et al.</em>, 2008</td>
</tr>
<tr>
<td>Victoria, Australia</td>
<td></td>
<td>X (verbal consent, written dissent)</td>
<td></td>
<td>Quinlivan &amp; Suriadi 2006</td>
</tr>
<tr>
<td>Bavaria, Germany</td>
<td></td>
<td>X (written)</td>
<td></td>
<td>Liebl <em>et al.</em>, 2002</td>
</tr>
<tr>
<td>France</td>
<td></td>
<td>X (written)</td>
<td></td>
<td>Farrieaux <em>et al.</em>, 2003; Dhondt <em>et al.</em>, 2005</td>
</tr>
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<td>Maryland, United States</td>
<td></td>
<td>X</td>
<td></td>
<td>Faden <em>et al.</em>, 1982</td>
</tr>
<tr>
<td>Netherlands</td>
<td></td>
<td>X</td>
<td></td>
<td>Detmar <em>et al.</em>, 2007</td>
</tr>
<tr>
<td>United Kingdom</td>
<td></td>
<td>X (written)</td>
<td></td>
<td>Laing &amp; McIntosh 2004; Nicholls, 2010</td>
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<tr>
<td>United States (majority of US states)</td>
<td>X</td>
<td></td>
<td></td>
<td>Hiller, 1997; Newson 2006</td>
</tr>
<tr>
<td>Taiwan</td>
<td></td>
<td>X</td>
<td>X (although the authors write that while consent is meant to be express for supplementary screening, in many clinics and hospitals this is conducted routinely without consent)</td>
<td>Huang <em>et al.</em>, 2005</td>
</tr>
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</table>
## Appendix B: List of Conditions on the NBS Panel in Ontario*

<table>
<thead>
<tr>
<th>Deficiency/Disorder Names</th>
<th>Abbreviations</th>
<th>Type of Condition</th>
<th>Approximate Occurrence in Ontario</th>
</tr>
</thead>
<tbody>
<tr>
<td>Congenital hypothyroidism</td>
<td>CH</td>
<td>Endocrine</td>
<td>1/3,000</td>
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<tr>
<td>Congenital adrenal hyperplasia</td>
<td>CAH</td>
<td>Endocrine</td>
<td>1/15,000</td>
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<td>Sickle cell disease</td>
<td>HB S/S</td>
<td>Hemoglobin</td>
<td></td>
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<td>HB S/C</td>
<td>Hemoglobin</td>
<td>1/400 in some populations</td>
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<td>S-beta thalassemia</td>
<td>HB S/A</td>
<td>Hemoglobin</td>
<td></td>
</tr>
<tr>
<td>Biotinidase</td>
<td>BIO</td>
<td>Other</td>
<td>1/60,000</td>
</tr>
<tr>
<td>Transferase deficient galactosemia</td>
<td>GALT</td>
<td>Other</td>
<td>1/60,000</td>
</tr>
<tr>
<td>Cystic fibrosis</td>
<td>CF</td>
<td>Other</td>
<td>1/3,600</td>
</tr>
<tr>
<td>Carnitine uptake defect (Carnitine transport defect)</td>
<td>CUD</td>
<td>Fatty Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>Long-chain hydroxyacyl-CoA dehydrogenase</td>
<td>LCHAD</td>
<td>Fatty Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>Medium-chain acyl-CoA dehydrogenase</td>
<td>MCAD</td>
<td>Fatty Acid Disorders</td>
<td>1/10,000</td>
</tr>
<tr>
<td>Trifunctional protein</td>
<td>TFP</td>
<td>Fatty Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>Very long-chain acyl-CoA dehydrogenase</td>
<td>VLCAD</td>
<td>Fatty Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>Glutaric acidemia type 1</td>
<td>GA-1</td>
<td>Organic Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>3-Hydroxy 3-methylglutaric aciduria (3-Hydrox 3-methylglutaryl-CoA lyase)</td>
<td>HMG</td>
<td>Organic Acid Disorders</td>
<td>Unknown as so rare</td>
</tr>
<tr>
<td>Isovaleric acidemia (Isovaleryl-CoA dehydrogenase)</td>
<td>IVA</td>
<td>Organic Acid Disorders</td>
<td>1/100,000 to 1/200,000</td>
</tr>
<tr>
<td>3-Methylcrotonyl-CoA carboxylase</td>
<td>3-MCC</td>
<td>Organic Acid Disorders</td>
<td>1/50,000</td>
</tr>
<tr>
<td>Methylmalonic acidemia (Vitamin B12 Disorders)</td>
<td>Cbl-A,B</td>
<td>Organic Acid Disorders</td>
<td>\ 1/50,000</td>
</tr>
<tr>
<td>Methylmalonic Acidemia (methylmalonyl-CoA mutase)</td>
<td>MUT</td>
<td>Organic Acid Disorders</td>
<td></td>
</tr>
<tr>
<td>Deficiency/Disorder Names</td>
<td>Abbreviations</td>
<td>Type of Condition</td>
<td>Approximate Occurrence in Ontario</td>
</tr>
<tr>
<td>--------------------------</td>
<td>---------------</td>
<td>-------------------</td>
<td>----------------------------------</td>
</tr>
<tr>
<td>Beta ketothiolase (mitochondrial acetoacetyl-CoA thiolase; short-chain ketoacyl thiolase; T2)</td>
<td>BKT</td>
<td>Organic Acid Disorders</td>
<td>Unknown as very rare</td>
</tr>
<tr>
<td>Propionic acidemia (Propionyl-CoA carboxylase)</td>
<td>PROP</td>
<td>Organic Acid Disorders</td>
<td>1/100,000</td>
</tr>
<tr>
<td>Multiple carboxylase (Holocarboxylase synthetase)</td>
<td>MCD</td>
<td>Organic Acid Disorders</td>
<td>1/90,000</td>
</tr>
<tr>
<td>Argininosuccinate acidemia</td>
<td>ASA</td>
<td>Amino Acid Disorders</td>
<td>1/70,000</td>
</tr>
<tr>
<td>Citrullinemia type 1 (Argininosuccinate synthetase)</td>
<td>CIT</td>
<td>Amino Acid Disorders</td>
<td>1/60,000</td>
</tr>
<tr>
<td>Homocystinuria (cystathionine beta synthase)</td>
<td>HCY</td>
<td>Amino Acid Disorders</td>
<td>1/200,000 to 1/300,000</td>
</tr>
<tr>
<td>Maple syrup urine disease (branched-chain ketoacid dehydrogenase)</td>
<td>MSUD</td>
<td>Amino Acid Disorders</td>
<td>1/200,000</td>
</tr>
<tr>
<td>Phenylketonuria/hyperphenylalaninemia</td>
<td>PKU</td>
<td>Amino Acid Disorders</td>
<td>1/12,000</td>
</tr>
<tr>
<td>Tyrosinemia Type 1</td>
<td>TYR-l</td>
<td>Amino Acid Disorders</td>
<td>1/100,000</td>
</tr>
</tbody>
</table>

*The information in this table was compiled from two sources: 1) The list of conditions screened for in Ontario, including the names, abbreviations, and types of conditions, was drawn from the Canadian Organization for Rare Disorders’s Newborn Screening in Canada Status Report, which was updated May 5, 2012 (Adams, 2012; http://raredisorders.ca/documents/CanadaNBSstatusupdatedMay52012.pdf). 2) The approximate occurrence of each condition on the screening panel in Ontario was drawn from Newborn Screening Ontario’s (n.d.i) website (http://www.newbornscreening.on.ca/bins/content_page.asp?cid=6-18&lang=1).
## Appendix C: Number of Conditions Screened for Across Canada

(This list includes the number of conditions currently *implemented* in each province/territory)

<table>
<thead>
<tr>
<th>Province/Territory</th>
<th>Core Conditions</th>
<th>Secondary Target Conditions</th>
<th>Total Number of Conditions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Newfoundland &amp; Labrador</td>
<td>5</td>
<td>1</td>
<td>6</td>
</tr>
<tr>
<td>Prince Edward Island</td>
<td>10</td>
<td>4</td>
<td>14</td>
</tr>
<tr>
<td>Nova Scotia</td>
<td>10</td>
<td>4</td>
<td>14</td>
</tr>
<tr>
<td>New Brunswick</td>
<td>10</td>
<td>4</td>
<td>14</td>
</tr>
<tr>
<td>Quebec*</td>
<td>12</td>
<td>3</td>
<td>15</td>
</tr>
<tr>
<td>Ontario</td>
<td>28</td>
<td>0</td>
<td>28</td>
</tr>
<tr>
<td>Manitoba</td>
<td>28</td>
<td>24</td>
<td>52</td>
</tr>
<tr>
<td>Saskatchewan</td>
<td>23</td>
<td>20</td>
<td>43</td>
</tr>
<tr>
<td>Alberta</td>
<td>19</td>
<td>0</td>
<td>19</td>
</tr>
<tr>
<td>British Columbia</td>
<td>22</td>
<td>8</td>
<td>30</td>
</tr>
<tr>
<td>Yukon</td>
<td>21</td>
<td>8</td>
<td>29</td>
</tr>
<tr>
<td>Northwest Territories</td>
<td>18</td>
<td>0</td>
<td>18</td>
</tr>
<tr>
<td>Nunavut (Kitimeot region)</td>
<td>18</td>
<td>0</td>
<td>18</td>
</tr>
<tr>
<td>Nunavut (Kivilliq region)</td>
<td>24</td>
<td>0</td>
<td>24</td>
</tr>
<tr>
<td>Nunavut (Baffin region)</td>
<td>11</td>
<td>3</td>
<td>14</td>
</tr>
</tbody>
</table>

---


* “Bloodspot expansion under review; 2nd screen by urine collected by parent at newborn Day 21: 12 conditions of amino, urea cycle & organic acids & 11 transport disorders of amino acids (Fanconi syndrome, Cystinurias, Hartnup syndrome, Cystathioninemia, Prolidase deficiency, etc.)” (Adams, 2012: 1).

** All regions except for Nunavut Kitimeot region offer hearing screening either universally or to select populations or by request (Adams, 2012: 1).
Appendix D: The Nuffield Council on Bioethics’ “The Intervention Ladder”

**Box 3.2: The Intervention Ladder**

The range of options available to government and policy makers can be thought of as a ladder of interventions, with progressive steps from individual freedom and responsibility towards state intervention as one moves up the ladder. In considering which ‘rung’ is appropriate for a particular public health goal, the benefits to individuals and society should be weighed against the erosion of individual freedom. Economic costs and benefits would need to be taken into account alongside health and societal benefits. The ladder of possible policy action is as follows:

<table>
<thead>
<tr>
<th>Level of Intervention</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>Eliminate choice</td>
<td>Regulate in such a way as to entirely eliminate choice, for example through compulsory isolation of patients with infectious diseases.</td>
</tr>
<tr>
<td>Restrict choice</td>
<td>Regulate in such a way as to restrict the options available to people with the aim of protecting them, for example removing unhealthy ingredients from foods, or unhealthy foods from shops or restaurants.</td>
</tr>
<tr>
<td>Guide choices through disincentives</td>
<td>Fiscal and other disincentives can be put in place to influence people not to pursue certain activities, for example through taxes on cigarettes, or by discouraging the use of cars in inner cities through charging schemes or limitations of parking spaces.</td>
</tr>
<tr>
<td>Guide choices through incentives</td>
<td>Regulations can be offered that guide choices by fiscal and other incentives, for example offering tax-breaks for the purchase of bicycles that are used as a means of travelling to work.</td>
</tr>
<tr>
<td>Guide choices through changing the default policy</td>
<td>For example, in a restaurant, instead of providing chips as a standard side dish (with healthier options available), menus could be changed to provide a more healthy option as a standard (with chips as an option available).</td>
</tr>
<tr>
<td>Enable choice</td>
<td>Enable individuals to change their behaviours, for example by offering participation in an NHS ‘stop smoking’ programme, building cycle lanes, or providing free fruit in schools.</td>
</tr>
<tr>
<td>Provide information</td>
<td>Inform and educate the public, for example as part of campaigns to encourage people to walk more or eat five portions of fruit and vegetables per day.</td>
</tr>
<tr>
<td>Do nothing or simply monitor the current situation</td>
<td></td>
</tr>
</tbody>
</table>

Appendix E: Towards a Framework for Thinking Through Least Restrictive Alternatives for Medical Public Health Interventions

Among the goals of public health ethics is to encourage individuals, programs, and governments engaged in public health initiatives to strive towards achieving public health goals using the least restrictive approaches possible (Upshur, 2002; Childress et al., 2002; Thompson et al., 2006; Baylis et al., 2008). The Nuffield Council on Bioethics (2007) created an “Intervention Ladder” to outline the host of options available to achieve public health objectives for non-medical public health interventions ranging from least restrictive to most restrictive approaches (see Appendix D). In considering medical public health interventions such as newborn screening, for example, informed consent, both express and implied, are arguably two least restrictive alternatives that can be used to achieve broader public health goals. However, attempting to superimpose such least restrictive alternatives for medical interventions onto the Nuffield Council on Bioethics’ “Intervention Ladder” for non-medical interventions raised a number of challenges. Moreover, given that medical public health interventions have distinct challenges, I created a preliminary framework to aid in thinking through the range of possible approaches that can be considered to achieve medical public health goals and objectives.  

First, I chose to present a handful of the various possible least restrictive approaches that could be implemented to achieve medical public health goals in terms of a spectrum rather than a ladder. I maintain that a spectrum demonstrates more clearly that individual autonomy and “state infringement” can be intertwined at either end of the spectrum depending on how well or poorly the approach is designed and/or executed. For example, “least restrictive alternatives” that seem ideal in theory but do little to foster individual autonomy in practice can reflect a certain level of restrictiveness or coercion. Conversely, well designed and executed public health measures that fall on the more restrictive side of the spectrum of approaches (with the exception of mandatory measures without exemption provisions) can potentially foster and respect individual autonomy if such measures are implemented in a transparent manner. In addition, given that least restrictive alternatives can cause harm (Macintyre & Petticrew, 2000; Nuffield Council on Bioethics, 2007; Guttman & Salmon, 2004; Faden & Shebaya, 2010), this preliminary framework provides a visual representation of such a possibility in order to serve as a reminder that least restrictive alternatives in theory should be evaluated for their effectiveness in practice.

The visual representation of this spectrum of autonomy within the context of medical public health interventions (see Figure 1 below) begins with the two poles frequently referenced in public health ethics scholarship: protecting individual rights and freedoms on the one hand and protecting population health on the other. Between these two poles I have placed seven possible approaches to address medical public health initiatives that reflect a gradation of intervention from least restrictive to most restrictive. The colour gradation endeavours to capture the spectrum of individual autonomy (and “state infringement”) across

76 I recognize that this is not a comprehensive list and that other approaches are perhaps worthy of inclusion. This figure is meant to serve as a point of departure for further discussion and debate.
these possibilities. I then offer a preliminary rationale for the specific placement of each of these options along the spectrum, fully recognizing that arguments can likely be made for alternative positioning. This figure serves as a point of departure for future discussion, deliberation, exploration, and refinement.

Figure 1: Autonomy Spectrum for Medical Public Health Interventions

<table>
<thead>
<tr>
<th>Protecting Individual Rights and Freedoms</th>
<th>Protecting Population Health</th>
</tr>
</thead>
<tbody>
<tr>
<td>Autonomy</td>
<td>State Infringement</td>
</tr>
</tbody>
</table>

Justification for Positioning Along the Autonomy Spectrum:

Express Consent
Express consent for a medical public health intervention, whether written or verbal, in theory offers individuals the most autonomy in terms of whether they wish to have the intervention done to them or their children.

Opt-In
An opt-in approach to a medical public health intervention is a least restrictive alternative, but is slightly more coercive than express consent. Depending on how the approach is implemented, challenges of opt-in programs focus on the fact that while individuals must make a conscious and ideally informed decision about whether to have the intervention they must first be aware that the intervention is available.
**Implied Consent**

Although implied consent is among the least restrictive options, it reflects more state infringement. While the language of consent is embedded in the concept of implied consent and that individuals in theory must be fully informed, if individuals happen not to be informed about the clinical public health intervention in question, they will have something happen to them whether they want it to or not or whether they fully understand the implications or not.

**Community Consent**

Community consent has been put forth as a possible least restrictive approach to consent for public health initiatives (Public Health Ontario, 2012). Considering community consent or community consensus as an approach to medical public health interventions suggests that informed consent has been factored into the approach in some way. However, like all approaches to consent the extent to which this approach is more or less coercive will likely have to do with how the community consent is executed. Questions that should be contemplated in the process of determining the moral justification of such an approach include, who is ultimately bestowed with the power to give such consent and how does that person (or people) decide whether to consent or not on behalf of the community? Another question related to this approach is whether a broader form of community outreach occurred in advance of the community consent. If so, who attended? Was it a representative sample of the community? Also, within the context of community consent, are individuals allowed to refuse? Are individuals informed that someone (e.g., their leader, elected official, etc.) has consented to a clinical public health intervention on their behalf? What level of transparency exists around the medical public health intervention in question? The specifics around how this particular approach to consent is implemented will likely influence the extent to which individual autonomy is considered within a more community based approach to a medical public health initiative.

**Opt-Out**

An opt-out approach to a clinical public health intervention is arguably closer to the community health pole than the individual rights and freedoms pole given that unless someone is aware of the intervention and that they have the ability to refuse the intervention, the clinical intervention in question may just happen to them and/or their child.

**Mandatory Intervention with Exemption Provisions**

With this approach, certain individuals would be allowed to be exempt from undergoing the intervention in question. However, mandatory approaches that have exemption clauses raise questions about the exemption criteria. Specifically, who is exempt from the intervention, what must individuals do in order to obtain/receive the exemption, who decides on the exemption criteria and/or whether someone is able to exempt, how are such decisions made, and are individuals aware that such exemptions exist (Sutton & Upshur, 2010)? The answers
to these questions will likely contribute to how coercive this approach is, recognizing that it falls on the more coercive side of the spectrum.

**Mandatory Intervention Without Exemption Provisions**
A mandatory clinical public health intervention without the possibility to opt-out epitomizes the full extent of the State’s police powers. In such scenarios receiving the intervention in question would be obligatory: no exceptions. Historically, failure to comply resulted in heavy fines and/or incarceration (Wynia, 2007).
What does the testing look for?

The testing done for the Ontario Newborn Screening Program looks for at least 28 disorders that can cause health problems in babies and children.

These disorders include:

**Metabolic disorders:** When the body is not able to break down (metabolize) certain substances in food like fats, proteins or sugars, they can accumulate in the body and cause serious health problems.

**Endocrine disorders:** The endocrine system, which is responsible for producing hormones, can sometimes produce too much or too little of some hormones, leading to sickness or developmental disabilities.

**Blood disorders:** Oxygen is carried through the body in the hemoglobin found in red blood cells. If hemoglobin is not formed or not working properly, red blood cells break down, leading to health problems including severe anemia and serious infections.

**Cystic Fibrosis:** A condition that causes mucus to build up in the lungs, digestive system and other organs.

Newborn Screening: A healthy start leads to a healthier life

Want to learn more?

For more information about the Ontario Newborn Screening Program, speak to your doctor or midwife. Visit the Program’s website at www.newbornscreening.on.ca, or call 1-866-532-3161 (TTY 1-800-387-5559).
Getting the best start.

As a new or expecting parent, your baby's health is important to you. To help your baby get the best start in life and stay healthy, your newborn—and every other newborn in Ontario—will be screened for at least 28 disorders.

Although most babies with these disorders look healthy at birth, they may be at risk of having serious health problems—including developmental disabilities, recurrent sickness and even death—if their disorder is not detected and treated. Early identification could save your baby’s life, and is the key to effective treatment.

Early detection leads to early treatment.

Individually, these disorders are rare. As a group, they will affect about 150 out of approximately 140,000 babies born each year in Ontario. Even if there is no family history of these disorders, your baby is still at risk. By testing newborns within the first days of their lives, disorders can be treated early, reducing the chance of serious health problems later in life.

This pamphlet will provide you with information about the Ontario Newborn Screening Program, and answer questions that you may have about the newborn screening test.

A small test, producing big benefits.

In order to perform the screening test, a small sample of blood is taken from your baby by pricking the heel. The blood is collected on a special paper card and then sent to the Newborn Screening Program’s laboratory for testing.

Blood samples can be taken anytime between one day (24 hours) and seven days after your baby is born. The best time to collect the blood sample is when your baby is between two days (48 hours) and three days (72 hours) old. If your baby is tested before one day (24 hours) of age, your baby’s health care provider should repeat the test within five days, at the baby’s first checkup.

Screening results: high risk and low risk.

A screening test only shows whether there is a high or low risk that your baby has a disorder. It is important to understand that the test does not make a diagnosis of a disorder, but only identifies babies who need further testing.

Once the laboratory has received and analyzed your baby’s blood sample, one of the following will occur:

1. Your baby screens negative for all the disorders. A report will be sent by mail to your hospital and/or health care provider. It will be filed in your baby’s medical records.

   More than 99 per cent of babies screened will receive a “screen negative” result. This means there is a very low risk that your baby has one of these disorders. On very rare occasions, the newborn screening test may miss a baby with one of these disorders.

   2. The laboratory may need another blood sample. It may be that the first sample was not taken properly, there wasn’t enough blood to complete the testing, or there was some other problem with the sample. In this case, your baby’s health care provider will contact you and arrange for another blood sample to be taken as soon as possible.

   3. Your baby screens positive for one of the conditions. A screen positive does not necessarily mean that your baby has a disorder, but only that further testing is needed. Your baby’s health care provider will contact you right away to make arrangements for follow-up at a hospital where specialists can do further testing. If a diagnosis of a disorder is made, the hospital will provide your baby with treatment and your family with counselling and advice.

The Ontario Newborn Screening Program also issues a report by mail to your hospital and/or health care provider, which will be filed in your baby’s medical records. It is important to remember that less than 1 per cent of babies tested will receive a “screen positive” result.
Appendix G: Parent Information Sheet

General Instructions:

1. **ALL** information on this form must be complete for identification and reporting purposes. Print identifying information on this form with pressure using a ball-point pen. If a stamp or label is used, stamp or label all copies.

2. Do not use this kit after expiry date (shown on filter paper).

3. Blood samples are to be taken on every infant before discharge from the hospital regardless of age, milk intake, or age at discharge.

4. Optimum time for screening is 48 to 72 hours after birth. Any infant discharged before the age of 24 hours must have a repeat test within 5 days.

Instructions for Blood Collection:

1. Warm infant’s heel with soft cloth moistened with warm water for 3 to 5 minutes.

2. Cleanse skin with alcohol only, let dry and puncture heel with 2.0 mm disposable sterile lancet (see diagram). Wipe away first blood drop with sterile gauze and allow another large blood drop to form.

3. Lightly touch the filter paper to the large blood drop while viewing from the other side to ensure complete penetration and saturation of the entire circle with a single application. Apply blood to one side of the filter paper only. It is preferable not to use capillary tube to transfer blood as this may damage filter paper.

4. Fill the remaining circles in the same manner. If blood flow is diminished, repeat steps 1 and 2. To enhance blood flow, very gentle intermittent pressure may be applied to area surrounding puncture site.

5. Allow blood on card to dry thoroughly for a minimum of three hours at room temperature in a horizontal position. Do not allow card to touch anything else while drying. Never superimpose one wet specimen collection paper on another before thoroughly drying. Do not place collection
Dear Parent,

A blood sample has been taken from your baby's heel to screen for serious diseases which can cause mental retardation, poor growth, or death if not treated. The pamphlet "Newborn Screening: A healthy start leads to a healthier life" describes this test. If you have not received this pamphlet, ask your nurse, baby's physician or your midwife for a copy.

If your baby's sample is taken before he or she is 24 hours old, ask your midwife or your baby's physician to repeat the newborn screening test within 5 days.

The Ontario Newborn Screening Program (ONSP) will report screening results to the hospital or health care provider who sent the sample. If a repeat sample is needed, the Program will notify the health care provider who submitted the sample. If the screen is positive, you or your baby's health care provider will be contacted directly and the ONSP physicians will refer your baby to specialists at a treatment centre. Personal health information will be shared between the health care providers involved in newborn screening and diagnosis to ensure your baby receives appropriate care and follow-up. You may prefer not to have this information shared, in which case, please make your wishes known and discuss this with your health care provider and/or contact the Ontario Newborn Screening Program.

If you have any questions or would like more information about the program, or how we use your health information, please speak to your baby's doctor, your midwife, contact the Ontario Newborn Screening Program or visit our website at www.newbornscreening.on.ca.

INSTRUCTIONS TO HOSPITAL:
Remove this top sheet and give to parent.

*Fill out the first copy of the requisition form.
Appendix H: Letters of Invitation

Date

Dear

My name is Erica Sutton and I am a PhD candidate at the Dalla Lana School of Public Health and the Joint Centre for Bioethics at the University of Toronto. I am conducting a case study on Ontario’s expanded newborn screening program under the supervision of Drs. Ross Upshur, Fiona Miller, and Patricia McKeever. The goal of my research is to document the expansion of Ontario’s expanded newborn screening program and identify the ethical principles and social, political, and economic factors that shaped, and will continue to shape, the program’s future.

Given your expertise, the purpose of this email is to ask if you would be willing to participate as a volunteer in this research study. The University of Toronto’s research ethics board has approved this study.

What is this study about?
This case study aims to situate Ontario’s expanded newborn screening program nationally and internationally. This study will also look at how Ontario’s newborn screening program came to be, explore how ethical issues are identified and considered in public health decision-making processes, and gain insight into the perceived future of expanded newborn screening.

Why is this study important?
As newborn screening technologies continue to develop, the possibilities for how the technologies can be used within Ontario’s public health program expand. Public health departments recognize the importance of identifying and addressing ethical issues associated with their programs. Understanding how key informants identify the various ethical challenges in expanded newborn screening is crucial to informing future policy and practice decisions.

Who is being approached to participate in this study?
Approximately 30 individuals are being asked to participate in this case study based on their past and/or present involvement in the expansion of Ontario’s newborn screening program. I hope to recruit individuals who represent a variety of perspectives including health care providers, influential activists, policy makers, government officials, non-governmental organizations, academics and representatives from the private sector. Because you have insight into and experience with Ontario’s newborn screening program, I am inviting you to participate in this study.

What would you have to do as a participant?
If you volunteer to be in this study you will be asked to participate in a one-on-one interview at a time that is convenient for you. The interview questions will ask you to talk about how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on newborn screening in Ontario and how you see the future of Ontario’s newborn screening program.

Thank you in advance for considering this request. Please feel welcome to contact me by email (erica.sutton@utoronto.ca) or by telephone (647-345-7711) if you are interested in participating or for further discussion. If at all possible I would appreciate hearing from you within the next two weeks.

Yours sincerely,

Erica Sutton

155 College Street, Suite 754, Toronto, Ontario Canada M5T 1P8 Tel.: [416] 978-2709 Fax: [416] 978-1911
www.utoronto.ca/jcb

A partnership among the University of Toronto; Baycrest Centre for Geriatric Care; Bloorview Kids Rehab; Centre for Addiction and Mental Health; Centre for Clinical Ethics, a joint venture of Providence Centre, St. Joseph’s Health Centre, and St. Michael’s Hospital; The Hospital for Sick Children; Humber River Regional Hospital; Mount Sinai Hospital; North York General Hospital; Sunnybrook Health Sciences Centre; Toronto Community Care Access Centre; Trillium Health Centre; Toronto Rehabilitation Institute; and University Health Network.
Dear

My name is Erica Sutton and I am a PhD candidate at the Dalla Lana School of Public Health and the Joint Centre for Bioethics at the University of Toronto. I am conducting a case study on Ontario’s expanded newborn screening program under the supervision of Drs. Ross Upshur, Fiona Miller, and Patricia McKeever. The goal of my research is to document the expansion of Ontario’s expanded newborn screening program and identify the ethical principles and social, political, and economic factors that shaped, and will continue to shape, the program’s future.

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What would you have to do as a participant?
If you volunteer to be in this study you will be asked to participate in a one-on-one interview at a time that is convenient for you. The interview questions will ask you to talk about how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on newborn screening in Ontario and how you see the future of Ontario’s newborn screening program.

Thank you in advance for considering this request. Please feel welcome to contact me by email (ericasutton@utoronto.ca) or by telephone (647-345-7711) if you are interested in participating or for further discussion. If at all possible I would appreciate hearing from you within the next two weeks.

If you have any concerns about your rights as a research subject please contact - Dr. Albert Clark, Chair of the Queen’s University Health Sciences and Affiliated Teaching Hospitals Research Ethics Board at (613) 533-6081.

Yours sincerely,

Erica Sutton

155 College Street, Suite 754, Toronto, Ontario Canada MST 1P8 Tel.: [416] 978-2709 Fax: [416] 978-1911

www.utoronto.ca/jcb

A partnership among the University of Toronto; Baycrest Centre for Geriatric Care; Bloorview Kids Rehab; Centre for Addiction and Mental Health; Centre for Clinical Ethics, a joint venture of Providence Centre, St. Joseph’s Health Centre, and St. Michael’s Hospital; The Hospital for Sick Children; Humber River Regional Hospital; Mount Sinai Hospital; North York General Hospital; Sunnybrook Health Sciences Centre; Toronto Community Care Access Centre; Trillium Health Centre; Toronto Rehabilitation Institute; and University Health Network.
Date

Dear

My name is Erica Sutton and I am a PhD candidate at the Dalla Lana School of Public Health and the Joint Centre for Bioethics at the University of Toronto. I am conducting a case study on Ontario’s expanded newborn screening program under the supervision of Drs. Ross Upshur, Patricia McKeever, and Fiona Miller at the University of Toronto and Dr. Jeff Nisker at the London Health Sciences Centre, Victoria Hospital. The goal of my research is to document the expansion of Ontario’s expanded newborn screening program and to identify the social, political, economic, and ethical principles that shaped, and will continue to shape, the program’s future.

Given your expertise, the purpose of this email is to request your cooperation as a voluntary participant in this research.

What is this study about?
This case study aims to situate Ontario’s expanded newborn screening program nationally and internationally. This study will also look at how Ontario’s newborn screening program came to be, explore how ethical issues are identified and considered in public health decision-making processes, and gain insight into the perceived future of expanded newborn screening.

Why is this study important?
As newborn screening technologies continue to develop, the possibilities for how the technologies can be used within Ontario’s public health program expand. Public health departments recognize the importance of identifying and addressing ethical issues associated with their programs. Understanding how key informants identify the various ethical challenges in expanded newborn screening is crucial to informing future policy and practice decisions.

Who is being approached to participate in this study?
Approximately 70 individuals are being asked to participate in this case study based on their past and/or present involvement in the expansion of Ontario’s newborn screening program. I hope to recruit individuals who represent a variety of perspectives including health care providers, influential activists, policy makers, government officials, non-governmental organizations, academics and representatives from the private sector. Because you have insight into and experience with Ontario’s newborn screening program, I am inviting you to participate in this study.

What would you have to do as a participant?
If you volunteer to be in this study you will be asked to participate in a one-on-one interview at a time that is convenient for you. The interview questions will ask you to talk about how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on newborn screening in Ontario and how you see the future of Ontario’s newborn screening program.

Thank you in advance for considering this request. Please feel welcome to contact me by email (ericasutton@utoronto.ca) or by telephone (647-345-7711) if you are interested in participating or for further discussion. If at all possible I would appreciate hearing from you within the next two weeks. If you have any concerns about your rights as a research subject please contact Dr. David Hill, Scientific Director, Lawson Health Research Institute at (519) 667-6649.

Yours sincerely,

Erica Sutton

155 College Street, Suite 754, Toronto, Ontario Canada M5T 1P8 Tel.: [416] 978-2709 Fax: [416] 978-1911
www.utoronto.ca/jcb

A partnership among the University of Toronto; Baycrest Centre for Geriatric Care; Bloorview Kids Rehab; Centre for Addiction and Mental Health; Centre for Clinical Ethics, a joint venture of Providence Centre, St. Joseph’s Health Centre, and St. Michael’s Hospital; The Hospital for Sick Children; Humber River Regional Hospital; Mount Sinai Hospital; North York General Hospital; Sunnybrook Health Sciences Centre; Toronto Community Care Access Centre; Trillium Health Centre; Toronto Rehabilitation Institute; and University Health Network.
Appendix I: Consent Forms

ONTARIO’S EXPANDED NEWBORN SCREENING PROGRAM: A CASE STUDY

INVESTIGATORS
Erica Sutton, Principal Investigator
Dalla Lana School of Public Health & Joint Centre for Bioethics, University of Toronto

Ross Upshur, Faculty Supervisor
Director of the Joint Centre for Bioethics, University of Toronto

SPONSORS/FUNDING
Canadian Institutes of Health Research
Health Care, Technology & Place
University of Toronto

INTRODUCTION
We invite you to take part in a research study on Ontario’s expanded newborn screening program. You have been asked to participate in this study because you have expertise in the influencing, shaping, decision-making, implementing, and/or the daily practice of this program.

First, we want you to know that:

Taking part in this case study research project is entirely voluntary.

You may choose not to take part, or you may withdraw from the study at any time. There are no negative consequences to not participating or to withdrawing from this study.

You may receive no benefit from taking part in this study. The research may give us knowledge that may help people in the future.

Now we will describe this research study. Before you decide to take part, please take as much time as you need to read the document and ask any questions you may have regarding this study.
WHY IS THIS STUDY BEING DONE?

The purpose of this study is to learn more about the evolution of Ontario’s expanded newborn screening program and the various individuals and organizations that contributed to its development. We hope to identify the social, political, economic, and ethical factors that shaped, and will continue to shape, the expansion of Ontario’s newborn screening program. Such knowledge is necessary given the growing national and international interest in newborn screening and the many potential uses for newborn blood spots. Also, public health departments recognize the importance of identifying and addressing the ethical issues associated with their programs.

WHAT IS INVOLVED IN THE STUDY?

- If you volunteer to be in this study you will be asked to participate in a one-on-one interview either in person or by telephone at a time that is convenient for you. We will also ask for your permission to speak with you again if we need to clarify or elaborate on certain comments that you made during the interview.

- If you decide to participate in this interview you will be asked questions regarding how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on expanded newborn screening in Ontario and where you see the future of newborn screening in Ontario heading.

- The interview will last between 60 and 90 minutes in length.

- Interviews will be tape recorded and transcribed.

WHAT ARE THE RISKS OF THE STUDY?

There are thought to be few risks of participating in this study, as you have been involved with Ontario’s newborn screening program in a professional capacity. However, if you are a parent who has/had a child with a rare genetic condition, participating in a research project might be different than speaking to government agencies and other organizations about your personal experience with newborn screening. Therefore, talking about some of these issues could make you upset.

ARE THERE BENEFITS TO TAKING PART IN THE STUDY?

There may be no benefit to you if you choose to take part in this study.

WHAT ARE MY OTHER OPTIONS?

You do not have to participate in this study if you do not want to. You may withdraw from the study at any time. There are no negative consequences of not participating in or withdrawing from this study.

Version Date: 08/10/2009
Ontario’s Expanded Newborn Screening Program: A Case Study

WHAT IF I CHANGE MY MIND?
You may stop participating in this study at any time simply by telling the coordinator of the study that you want to stop. You can even stop in the middle of the interview. If you decide to stop you can choose whether you want your interview data destroyed or not.

WHO ELSE WILL KNOW THAT I AM IN THIS STUDY?

- Case study research requires that names of people, places, and institutions are used to describe the specific facts of a case in order to tell the story. Therefore, we are asking for your permission to identify you by name when referring to specific facts related to the expansion of Ontario’s expanded newborn screening program.

(For example, if you were part of an influential committee and/or organization, or contributed to a critical policy, or wrote/helped write an important document that had an impact on the development of the program, and/or voted on issues related to newborn screening, we would describe your involvement and note your contributions by name).

You are free to decline this request and still participate in this study.

- We will not use your name when referring to your thoughts, opinions, and/or ideas about the past, present or future state of Ontario’s expanded newborn screening program. Pseudonyms or professional categories (e.g. government agency, non-profit organization, health care professional etc.) will be used to convey such information.

- All of the data we collect will be kept secure in locked cabinets and password protected computers.

- Only the researchers working on this study will be able to see what you said during the interview.

WILL I RECEIVE PAYMENT FOR BEING IN THIS STUDY?
You will not receive any payment for taking part in this study. However, if you travelled to participate in the interview your travel costs will be reimbursed.

WILL THE RESULTS OF THIS STUDY BE PUBLISHED?
Results from this research project will hopefully be published in scholarly journals. Researchers will also give public presentations based on the findings of this study. The researchers will provide you with published articles upon request.

WHO SHOULD I CONTACT IF I HAVE ANY QUESTIONS?
If you have any questions or problems regarding your participation in this study please contact Erica Sutton, the principal investigator and interviewer, at 647-345-7711 or by email at erica.sutton@utoronto.ca. You may also contact Dr. Ross Upshur, the faculty supervisor of this research at (416) 978-4756 or ross.upshur@sunnybrook.ca. You may also contact Rachel Zand, Director of the Office of Research Ethics at (416) 946-3273.

Version Date: 08/10/2009
CONSENT AND SIGNATURES

I have had the opportunity to discuss this study and my questions have been answered to my satisfaction. I consent to take part in the study with the understanding that I may withdraw at any time without any consequences. I have received a signed copy of this consent form.

I consent to having my name associated with facts related to the development of Ontario’s expanded newborn screening program: Y ☐ N ☐

I consent to a follow-up interview in the event that clarification or elaboration is necessary: Y ☐ N ☐

I voluntarily consent to participate in this study and continue with the interview.

______________________________  ______________________________
Signature of Participant                  Date

________________________
Printed Name

I believe that the person signing this form understands what is involved in the study and voluntarily agrees to participate.

______________________________  ______________________________
Signature of Investigator/Designee                  Date

Version Date: 08/10/2009
ONTARIO’S EXPANDED NEWBORN SCREENING PROGRAM: A CASE STUDY

INVESTIGATORS
Erica Sutton, Principal Investigator
Dalla Lana School of Public Health & Joint Centre for Bioethics, University of Toronto

Ross Upshur, Faculty Supervisor
Director of the Joint Centre for Bioethics, University of Toronto

SPONSORS/FUNDING
Canadian Institutes of Health Research
Health Care, Technology & Place
University of Toronto

INTRODUCTION
We invite you to take part in a research study on Ontario’s expanded newborn screening program. You have been asked to participate in this study because you have expertise in the influencing, shaping, decision-making, implementing, and/or the daily practice of this program.

First, we want you to know that:

Taking part in this case study research project is entirely voluntary.

You may choose not to take part, or you may withdraw from the study at any time. There are no negative consequences to not participating or to withdrawing from this study.

You may receive no benefit from taking part in this study. The research may give us knowledge that may help people in the future.

Now we will describe this research study. Before you decide to take part, please take as much time as you need to read the document and ask any questions you may have regarding this study.
**Why is this study being done?**

The purpose of this study is to learn more about the evolution of Ontario’s expanded newborn screening program and the various individuals and organizations that contributed to its development. We hope to identify the social, political, economic, and ethical factors that shaped, and will continue to shape, the expansion of Ontario’s newborn screening program. Such knowledge is necessary given the growing national and international interest in newborn screening and the many potential uses for newborn blood spots. Also, public health departments recognize the importance of identifying and addressing the ethical issues associated with their programs.

**What is involved in the study?**

- If you volunteer to be in this study you will be asked to participate in a one-on-one interview either in person or by telephone at a time that is convenient for you. We will also ask for your permission to speak with you again if we need to clarify or elaborate on certain comments that you made during the interview.

- If you decide to participate in this interview you will be asked questions regarding how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on expanded newborn screening in Ontario and where you see the future of newborn screening in Ontario heading.

- The interview will last between 60 and 90 minutes in length.

- Interviews will be tape recorded and transcribed.

**What are the risks of the study?**

There are thought to be few risks of participating in this study, as you have been involved with Ontario’s newborn screening program in a professional capacity. However, if you are a parent who has/had a child with a rare genetic condition, participating in a research project might be different than speaking to government agencies and other organizations about your personal experience with newborn screening. Therefore, talking about some of these issues could make you upset.

**Are there benefits to taking part in the study?**

There may be no benefit to you if you choose to take part in this study.

**What are my other options?**

You do not have to participate in this study if you do not want to. You may withdraw from the study at any time. There are no negative consequences of not participating in or withdrawing from this study.

**What if I change my mind?**

Version Date: 30/04/2010
You may stop participating in this study at any time simply by telling the coordinator of the study that you want to stop. You can even stop in the middle of the interview. If you decide to stop you can choose whether you want your interview data destroyed or not.

**WHO ELSE WILL KNOW THAT I AM IN THIS STUDY?**

- Case study research requires that names of people, places, and institutions are used to describe the specific facts of a case in order to tell the story. Therefore, we are asking for your permission to identify you by name when referring to specific facts related to the expansion of Ontario’s expanded newborn screening program.

  (For example, if you were part of an influential committee and/or organization, or contributed to a critical policy, or wrote/helped write an important document that had an impact on the development of the program, and/or voted on issues related to newborn screening, we would describe your involvement and note your contributions by name).

You are free to decline this request and still participate in this study.

- We will not use your name when referring to your thoughts, opinions, and/or ideas about the past, present or future state of Ontario’s expanded newborn screening program. Pseudonyms or professional categories (e.g. government agency, non-profit organization, health care professional etc.) will be used to convey such information.

- All of the data we collect will be kept secure in locked cabinets and password protected computers.

- Only the researchers working on this study will be able to see what you said during the interview.

**WILL I RECEIVE PAYMENT FOR BEING IN THIS STUDY?**

You will not receive any payment for taking part in this study. However, if you travelled to participate in the interview your travel costs will be reimbursed.

**WILL THE RESULTS OF THIS STUDY BE PUBLISHED?**

Results from this research project will hopefully be published in scholarly journals. Researchers will also give public presentations based on the findings of this study. The researchers will provide you with published articles upon request.

**WHO SHOULD I CONTACT IF I HAVE ANY QUESTIONS?**

If you have any questions or problems regarding your participation in this study please contact Erica Sutton, the principal investigator and interviewer, at 647-345-7711 or by email at erica.sutton@utoronto.ca. You may also contact Dr. Ross Upshur, the faculty supervisor of this research at (416) 978-4756 or ross.upshur@sunnybrook.ca. If you have any concerns about your rights as a research subject please contact - Dr. Albert Clark, Chair of the Queen’s University Health Sciences and Affiliated Teaching Hospitals Research Ethics Board at (613) 533-6081.
CONSENT AND SIGNATURES

I have had the opportunity to discuss this study and my questions have been answered to my satisfaction. I consent to take part in the study with the understanding that I may withdraw at any time without any consequences. I have received a signed copy of this consent form.

I consent to having my name associated with facts related to the development of Ontario’s expanded newborn screening program: Y [ ] N [ ]

I consent to a follow-up interview in the event that clarification or elaboration is necessary: Y [ ] N [ ]

I voluntarily consent to participate in this study and continue with the interview.

__________________________________________________________
Signature of Participant                                      Date

__________________________________________________________
Printed Name

I believe that the person signing this form understands what is involved in the study and voluntarily agrees to participate.

__________________________________________________________
Signature of Investigator/Designee                            Date

Version Date: 30/04/2010
ONTARIO’S EXPANDED NEWBORN SCREENING PROGRAM: A CASE STUDY

INVESTIGATORS

Erica Sutton, Student Researcher
Dalla Lana School of Public Health & Joint Centre for Bioethics, University of Toronto

Ross Upshur, Faculty Supervisor
Director of the Joint Centre for Bioethics, University of Toronto

Jeff Nisker, Principal Investigator
Professor of Obstetrics-Gynecology & Oncology,
London Health Sciences Centre, Victoria Hospital

SPONSORS/FUNDING

Canadian Institutes of Health Research
Health Care, Technology & Place
University of Toronto

INTRODUCTION

We invite you to take part in a research study on Ontario’s expanded newborn screening program. You have been asked to participate in this study because you have expertise in the influencing, shaping, decision-making, implementing, and/or the daily practice of this program. A number of individuals from the London Health Sciences Centre have been purposively invited to participate in this study and approximately 45 other individuals from health care institutions, government agencies and non-government organizations across the province have been invited to participate as well.

First, we want you to know that:

Taking part in this case study research project is entirely voluntary.

You may choose not to take part, or you may withdraw from the study at any time. There are no negative consequences to not participating or to withdrawing from this study.

You may receive no benefit from taking part in this study. The research may give us knowledge that may help people in the future.

Now we will describe this research study. Before you decide to take part, please take as much time as you need to read the document and ask any questions you may have regarding this study.
Why is this study being done?

The purpose of this study is to learn more about the evolution of Ontario’s expanded newborn screening program and the various individuals and organizations that contributed to its development. We hope to identify the social, political, economic, and ethical factors that shaped, and will continue to shape, the expansion of Ontario’s newborn screening program. Such knowledge is necessary given the growing national and international interest in newborn screening and the many potential uses for newborn blood spots. Also, public health departments recognize the importance of identifying and addressing the ethical issues associated with their programs.

What is involved in the study?

- If you volunteer to be in this study you will be asked to participate in a one-on-one interview either in person or by telephone at a time that is convenient for you. We will also ask for your permission to speak with you again if we need to clarify or elaborate on certain comments that you made during the interview.

- If you decide to participate in this interview you will be asked questions regarding how your work was or is related to the development, implementation, and/or practice of expanded newborn screening in Ontario. You will also be asked to discuss your views on expanded newborn screening in Ontario and where you see the future of newborn screening in Ontario heading.

- The interview will last between 60 and 90 minutes in length.

- Interviews will be tape recorded and transcribed.

What are the risks of the study?

There are thought to be few risks of participating in this study, as you have been involved with Ontario’s newborn screening program in a professional capacity. However, if you are a parent who has/had a child with a rare genetic condition, participating in a research project might be different than speaking to government agencies and other organizations about your personal experience with newborn screening. Therefore, talking about some of these issues could make you upset.

Are there benefits to taking part in the study?

There may be no benefit to you if you choose to take part in this study.

What are my other options?

You do not have to participate in this study if you do not want to. You may withdraw from the study at any time. There are no negative consequences of not participating in or withdrawing from this study.

What if I change my mind?

You may stop participating in this study at any time simply by telling the coordinator of the study that you want to stop. You can even stop in the middle of the interview. If you decide to stop you can choose whether you want your interview data destroyed or not.

Version Date: 19/07/2010
WHO ELSE WILL KNOW THAT I AM IN THIS STUDY?

- Case study research requires that names of people, places, and institutions be used to describe the specific facts of a case in order to tell the story. Therefore, we are asking for your permission to identify you by name when referring to specific facts related to the expansion of Ontario’s expanded newborn screening program.

(For example, if you were part of an influential committee and/or organization, or contributed to a critical policy, or wrote/helped write an important document that had an impact on the development of the program, and/or voted on issues related to newborn screening, we would describe your involvement and note your contributions by name).

You are free to decline this request and still participate in this study.

- On page 4 of this letter there are a series of check boxes. If you would like to be identified by name in this study please mark the box with the “Y” for “yes.” If you would prefer not to have your name mentioned please mark the box with the “N” for “no.” If you agree to being identified by name you will have an opportunity to read and make changes to how your name and contribution to the Ontario Newborn Screening Program will appear in future publications resulting from this research.

- We will not use your name when referring to your thoughts, opinions, and/or ideas about the past, present or future state of Ontario’s expanded newborn screening program. Pseudonyms or professional categories (e.g. government agency, non-profit organization, health care profession etc.) will be used to convey such information.

- All of the data we collect will be kept in locked cabinets and password-protected computers; only the researchers working on this study will be able to see what you said during the interview.

WILL I RECEIVE PAYMENT FOR BEING IN THIS STUDY?

You will not receive any payment for taking part in this study. However, if you travelled to participate in the interview your travel costs will be reimbursed.

WILL THE RESULTS OF THIS STUDY BE PUBLISHED?

Results from this research project will hopefully be published in scholarly journals. Researchers will also give public presentations based on the findings of this study. The researchers will provide you with published articles upon request.

WHO SHOULD I CONTACT IF I HAVE ANY QUESTIONS?

If you have any questions or problems regarding your participation in this study please contact Erica Sutton, the student researcher and interviewer at (647-345-7711) or by email at erica.sutton@utoronto.ca. You may also contact Dr. Jeff Nisker, the principal investigator of this research at (519) 685-8781 or jeff.nisker@lhsc.on.ca. If you have any concerns about your rights as a research subject please contact Dr. David Hill, Scientific Director, Lawson Health Research Institute at (519) 667-6649.

Version Date: 19/07/2010
CONSENT AND SIGNATURES

I have read the Letter of Information, have had the nature of the study explained to me and I agree to participate. All questions have been answered to my satisfaction. I have received a signed copy of this consent form.

I consent to having my name associated with facts related to the development of Ontario’s expanded newborn screening program: Y ☐ N ☐

I consent to a follow-up interview in the event that clarification or elaboration is necessary: Y ☐ N ☐

I voluntarily consent to participate in this study and continue with the interview.

________________________________________  ________________________________
Signature of Participant                      Date

________________________________________
Printed Name

I believe that the person signing this form understands what is involved in the study and voluntarily agrees to participate.

________________________________________  ________________________________
Signature of Investigator/Designee            Date

Version Date: 19/07/2010
Appendix J: Interview Guide

1. About You
   a. Could you please explain a bit about your involvement in Ontario’s newborn screening program as it relates to your professional and personal background.

2. Goals of NBS
   a. I am wondering what you think the goals of NBS are.
   b. Do you think these goals are or should be changing?
   c. Are the benefits of NBS different now than they were before the program expanded?
   d. Are the beneficiaries of NBS different now than they were?
   e. Are the risks of NBS different now than they were?
   f. Are the risks of NBS borne by different people now than they were?

3. Most Pressing Issues
   a. From your perspective, what would you say are some of the issues currently facing Ontario’s NBS program?
   b. Would you consider any of these as ethical concerns and why?

4. Hoped for Change
   a. If you could change anything in Ontario’s NBS program, what would it be?
      i. Is this a foreseeable change? Why or why not?

Now I want to shift gears a bit, and get your reaction to 5 or 6 issues that tend to dominate the literature addressing newborn screening, particularly the ethical issues related to NBS.

5. Informed Consent
   a. What are your views on the role of parental discretion in NBS?
   b. Did Ontario look at other models of consent? Why did it choose implied consent?
   c. Is the implied consent model the right model? What model should we use and why?
6. **Sample Storage and Research (and other secondary uses)**

   a. What are your views on the storage and secondary uses of blood spot samples? Benefits/Risks/Who etc.

   b. Do you view storage as part of Ontario’s NBS program? What about research?

   c. What do you think about the information that is being provided to parents regarding storage and research.

   d. Epidemiological research is mentioned in the FAQ of the NBS website. Can you speak to your awareness of non-anonymized research, is that currently occurring in Ontario or on the horizon?

7. **Carrier Status**

   a. What are your views on the use of disclosing carrier status information and other incidental findings to parents?

_Closing Questions_

Is there anyone you think I should talk to who could shed light on these issues?

Is there any literature you think I should read on these matters?

Is there anything else you would like to add?
Appendix K: Research Ethics Board Approval Letters

University of Toronto
Office of the Vice-President, Research
Office of Research Ethics

PROTOCOL REFERENCE #24435

October 26, 2009

Dr. Ross Upshur  Ms. Erica Sutton
Joint Centre for Bioethics  Joint Centre for Bioethics
88 College St.  88 College St.
Toronto, ON  Toronto, ON
M5G 1L4  M5G 1L4

Dear Dr. Upshur and Ms. Sutton,

Re: Your research protocol entitled “Ontario’s Expanded Newborn Screening Program: A Case Study”

ETHICS APPROVAL

Original Approval Date: October 26, 2009
Expiry Date: October 25, 2010
Continuing Review Level: 2

We are writing to advise you that a member of the Health Science Research Ethics Board has granted approval to the above-named research study, for a period of one year, under the REB’s delegated review process. Please ensure that you submit an Annual Renewal Form or a Study Completion Report at least 30 days prior to the expiry date of your study.

All your most recently submitted documents have been approved for use in this study.

Any changes to the approved protocol or consent materials must be reviewed and approved through the amendment process prior to its implementation. Any adverse or unanticipated events should be reported to the Office of Research Ethics as soon as possible.

If your research has funding attached, please contact the relevant Research Funding Officer in Research Services to ensure that your funds are released.

Best wishes for the successful completion of your project.

Yours sincerely,

Daniel Gyewu
Research Ethics Coordinator
Dear Dr. Upshur and Ms. Sutton,

Re: Your research protocol entitled, “Ontario’s Expanded Newborn Screening Program: A Case Study” by Dr. R. Upshur (supervisor), Ms. E. Sutton (PhD candidate)

We are writing to advise you that a member of the Health Sciences Research Ethics Board has granted approval to an amendment (received May 21, 2010) to the above referenced research study under the REB’s delegated review process. This amendment will allow for increasing the sample size from approximately 25 to 70 key informants to interview; the consent form offers participants the opportunity to consent to having their name linked to their role in ONSP; also a Permission for Publication form has been developed that will serve to document that each participant has had a chance to read the portions of the thesis that mention them by name and to make any changes.

All your most recently submitted documents have been approved for use in this study.

Any changes to the approved protocol or consent materials must be reviewed and approved through the amendment process prior to its implementation. Any adverse or unanticipated events should be reported to the Office of Research Ethics as soon as possible.

Best wishes for the successful completion of your project.

Yours sincerely,

Marianna Richardson
Research Ethics Coordinator
QUEEN'S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD

May 4, 2010

This Ethics Application was subject to:

☐ Full Board Review
☐ Meeting Date:
☒ Expedited Review

Ms. Erica Sutton
University of Toronto
Joint Centre for Bioethics
88 College Street
Toronto, ON M5G 1L4

Dear Ms. Sutton,

Study Title: Ontario's Expanded Newborn Screening Program: A Case Study
Co-Investigators: Dr. Ross Upshur

I am writing to acknowledge receipt of your recent ethics submission. We have examined the protocol and consent form for your project (as stated above) and consider it to be ethically acceptable. This approval is valid for one year from the date of the Chair's signature below. This approval will be reported to the Research Ethics Board. Please attend carefully to the following list of ethics requirements you must fulfill over the course of your study:

➢ Reporting of Amendments: If there are any changes to your study (e.g. consent, protocol, study procedures, etc.), you must submit an amendment to the Research Ethics Board for approval. (see http://www.queensu.ca/vpr/reb.htm).

➢ Reporting of Serious Adverse Events: Any unexpected serious adverse event occurring locally must be reported within 2 working days or earlier if required by the study sponsor. All other serious adverse events must be reported within 15 days after becoming aware of the information.

➢ Reporting of Complaints: Any complaints made by participants or persons acting on behalf of participants must be reported to the Research Ethics Board within 7 days of becoming aware of the complaint. Note: All documents supplied to participants must have the contact information for the Research Ethics Board.

➢ Annual Renewal: Prior to the expiration of your approval (which is one year from the date of the Chair's signature below), you will be reminded to submit your renewal form along with any new changes or amendments you wish to make to your study. If there have been no major changes to your protocol, your approval may be renewed for another year.

Yours sincerely,

[Signature]
Chair, Research Ethics Board

May 4, 2010

ORIGINAL TO INVESTIGATOR - COPY TO DEPARTMENT HEAD - COPY TO HOSPITALS / PAT (IF APPLICABLE) - FILE COPY

Study Code: MISC-140-10

➢ Investigators please note that if your trial is registered by the sponsor, you must take responsibility to ensure that the registration information is accurate and complete.
QUEEN'S UNIVERSITY HEALTH SCIENCES & AFFILIATED TEACHING HOSPITALS RESEARCH ETHICS BOARD

The membership of this Research Ethics Board complies with the membership requirements for Research Ethics Boards as defined by the Tri-Council Policy Statement; Part C Division 5 of the Food and Drug Regulations, OHRP, and U.S DHHS Code of Federal Regulations Title 45, Part 46 and carries out its functions in a manner consistent with Good Clinical Practices.

Federalwide Assurance Number: #FWA00004184
#IRB00001173

Current 2010 membership of the Queen's University Health Sciences & Affiliated Teaching Hospitals Research Ethics Board

Dr. A.F. Clark  Emeritus Professor, Department of Biochemistry, Faculty of Health Sciences, Queen's University (Chair)
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Dr. B. Simchison  Assistant Professor, Department of Anesthesiology, Queen's University
Dr. A.N. Singh  WHO Professor in Psychosomatic Medicine and Psychopharmacology Professor of Psychiatry and Pharmacology Chair and Head, Division of Psychopharmacology, Queen's University Director & Chief of Psychiatry, Academic Unit, Quinte Health Care, Belleville General Hospital
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Dr. S. Wood  Director, Office of Research Services (Ex-Officio)
Office of Research Ethics
The University of Western Ontario
Room 4180 Support Services Building, London, ON, Canada N6A 5C1
Telephone: (519) 661-3036 Fax: (519) 650-2466 Email: ethics@uwo.ca
Website: www.uwo.ca/research/ethics

Use of Human Subjects - Ethics Approval Notice

Principal Investigator: Dr. J. Nixter
Review Number: 17264E

Review Date: July 16, 2010
Review Level: Expedited
Approved Local # of Participants: 5

Protocol Title: Ontario's Expanded Newborn Screening Program: A Case Study
Department and Institution: Obstetrics & Gynaecology, London Health Sciences Centre
Sponsor: CIHR-CANADIAN INSTITUTE OF HEALTH RESEARCH

Ethics Approval Date: August 04, 2010
Expiry Date: June 30, 2011

Documents Reviewed and Approved: University of Toronto Protocol, Letter of Information and Consent, Invitation letter

Documents Received for Information: University of Toronto REB Approval, #24435

This is to notify you that the University of Western Ontario Research Ethics Board for Health Sciences Research Involving Human Subjects (HSREB) which is organized and operates according to the Tri-Council Policy Statement: Ethical Conduct of Research Involving Humans and the Health Canada/ICH Good Clinical Practice Practices: Consolidated Guidelines, and the applicable laws and regulations of Ontario has reviewed and granted approval to the above referenced study on the approval date noted above. The membership of this REB also complies with the membership requirements for REBs as defined in Division 5 of the Food and Drug Regulations.

The ethics approval for this study shall remain valid until the expiry date noted above assuming timely and acceptable responses to the HSREB’s periodic requests for surveillance and monitoring information. If you require an updated approval notice prior to that time you must request it using the UWO Updated Approval Request Form.

During the course of the research, no deviations from, or changes to, the protocol or consent form may be initiated without prior written approval from the HSREB except when necessary to eliminate immediate hazards to the subject or when the change(s) involve only logistical or administrative aspects of the study (e.g., change of monitor, telephone number). Expedited review of minor change(s) in ongoing studies will be considered. Subjects must receive a copy of the signed information/consent documentation.

Investigators must promptly also report to the HSREB:

a) changes increasing the risk to the participant(s) and/or affecting significantly the conduct of the study;
b) all adverse and unexpected experiences or events that are both serious and unexpected;
c) new information that may adversely affect the safety of the subjects or the conduct of the study.

If these changes/adverse events require a change to the information/consent documentation, and/or recruitment advertisement, the newly revised information/consent documentation, and/or advertisement, must be submitted to this office for approval.

Members of the HSREB who are named as investigators in research studies, or declare a conflict of interest, do not participate in discussion related to, nor vote on, such studies when they are presented to the HSREB.

Chair of HSREB: Dr. Joseph Gilbert
FDA Ref. #: IRB 00000940

Ethics Officer to Contact for Further Information

☐ Janice Sutherland (jsuther@uwo.ca)
☐ Elizabeth Wambolt (ewambolt@uwo.ca)
☐ Grace Kelly (grace.kelly@uwo.ca)
☐ Denise Grafton (dgraffon@uwo.ca)

This is an official document. Please retain the original in your files.
RESEARCH OFFICE REVIEW NO.: R-10-478

PROJECT TITLE: Ontario's Expanded Newborn Screening Program: A Case Study

PRINCIPAL INVESTIGATOR: Dr. Jeff Nisker

DATE OF REVIEW BY CRIC: September 9, 2010

Health Sciences REB#: 17264E

Please be advised that the above project was reviewed by the Clinical Research Impact Committee and the project:

Was Approved

PLEASE INFORM THE APPROPRIATE NURSING UNITS, LABORATORIES, ETC. BEFORE STARTING THIS PROTOCOL. THE RESEARCH OFFICE NUMBER MUST BE USED WHEN COMMUNICATING WITH THESE AREAS.

Dr. David Hill
V.P. Research
Lawson Health Research Institute

All future correspondence concerning this study should include the Research Office Review Number and should be directed to Sherry Paiva, CRIC Liaison, LHSC, Rm. C210, Nurses Residence, South Street Hospital.

cc: Administration