Title: Reconciling ethical and economic conceptions of value in health policy using the capabilities approach: A qualitative investigation of Non-Invasive Prenatal Testing

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Abstract

When evaluating new morally complex health technologies, policy decision-makers consider a broad range of different evaluations, which may include the technology’s clinical effectiveness, cost effectiveness, and social or ethical implications. This type of holistic assessment is challenging, because each of these evaluations may be grounded in different and potentially contradictory assumptions about the technology’s value.

One such technology where evaluations conflict is Non-Invasive Prenatal Testing (NIPT). Cost-effectiveness evaluations of NIPT often assess NIPT’s ability to deliver on goals (i.e. preventing the birth of children with disabilities) that social and ethical analyses suggest it should not have. Thus, cost effectiveness analyses frequently contradict social and ethical assessments of NIPT’s value.

We use the case of NIPT to explore how economic evaluations using a capabilities approach may be able to capture a broader, more ethical view of the value of NIPT. The capabilities approach is an evaluative framework which bases wellbeing assessments on a person’s abilities, rather than their expressed preferences. It is linked to extra-welfarist approaches in health economic assessment. Beginning with Nussbaum’s capability framework, we conducted a directed qualitative content analysis of interview data collected in 2014 from 27 Canadian women with personal experience of NIPT. We found that eight of Nussbaum’s ten capabilities related to options, states, or choices that women valued in the context of NIPT, and identified one new capability. Our findings suggest that women value NIPT for its ability to provide more and different choices in the prenatal care pathway, and that a capabilities approach...

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69 can indeed capture the value of NIPT in a way that goes beyond measuring health outcomes of ambiguous social and ethical value. More broadly, the capabilities approach may serve to resolve contradictions between ethical and economic evaluations of health technologies, and contribute to extra-welfarist approaches in the assessment of morally complex health technologies.

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1. Introduction

Non-invasive prenatal testing (NIPT) is a novel and morally challenging technology; one that raises ethical questions broader than NIPT itself, and evokes issues relevant to groups beyond those who interact with the technology directly (Hofmann, 2008). When making decisions about whether and how to implement this technology, policy decision-makers must consider a broad range of issues beyond clinical efficiency, including the technology’s economic, social, and ethical implications. This holistic policy consideration is complex and value-laden, and sometimes results in conflicting assessments (Giacomini et al., 2013). For example, with NIPT, economic evaluations often rely on assumptions that conflict with ethical analyses, resulting in assessments that raise ethical concerns about the technology’s use. In this research, we propose a theoretical approach that may help ameliorate these issues. We use NIPT as a case study to explore whether the capabilities approach could be used to resolve contradictions between economic and ethical framings of ‘value’ for morally challenging health technologies.

Non-invasive prenatal testing

Non-invasive prenatal testing (NIPT) analyzes cell-free fetal DNA circulating in maternal blood in order to gain information about the fetal genotype (Hui & Bianchi, 2017). This technology became commercially available in the United States, Canada, and Western Europe as early as 2011, and is now available globally (Chandrasekharan et al., 2014). In Canada, where this study takes place, NIPT was $800-1000 CAD when first introduced (Vanstone et al., 2015a); and is currently available for approximately $500 CAD (Nshimyumukiza et al., 2017). NIPT is currently used to detect trisomies 13, 18, 21, and sex chromosome abnormalities (ACOG, 2015),
but it is likely that NIPT will eventually be expanded to include a variety of genetic conditions (Hui & Bianchi, 2017).

NIPT is unique among the array of prenatal screening technologies currently available because it can provide information about the fetal genotype as early as 9 weeks’ gestation, with higher accuracy than existing screening tests and no risk of miscarriage (Vanstone et al., 2014). At this point, NIPT is still a screening test, and most clinical practice guidelines recommend that positive results be confirmed with invasive diagnostic tests (e.g. amniocentesis) which carry a small risk of miscarriage (ACOG, 2015).

NIPT has been rapidly and broadly adopted for prenatal genetic testing, as both a first-tier screening test for disability and as a second-tier screen to reduce the risk of iatrogenic miscarriage from invasive diagnostic procedures (Minear et al., 2015). This expansion has been facilitated by industry imperatives. NIPT technologies were developed by a number of different private companies, and in many places remain private-pay technologies, although some jurisdictions have recently offered coverage through public and private insurers (Minear et al., 2015; Vanstone et al., 2015b).

The commercial proliferation of NIPT preceded careful policy decision-making about its use and value (Vanstone et al., 2014). Evidence around NIPT’s clinical utility for different conditions and patient populations is still developing, and it is not yet clear how NIPT will integrate with existing prenatal testing technologies (Murdoch et al., 2017). The rapid expansion of NIPT has raised ethical concerns about the routinization of testing and erosion of informed decision-making processes (Deans & Newson, 2012; Lewis et al., 2013). Like other prenatal testing technologies, NIPT is challenged by ambiguously defined social and medical purposes.
Furthermore, ethical and policy analyses of NIPT are complicated because NIPT is not a homogeneous entity; it can refer to a number of slightly different methods for analyzing fetal genetic material, and is used by people and practitioners in different ways and for different purposes (Vanstone et al., 2015b).

**Economic and Ethical Evaluations of NIPT**

Policy evaluations of health technology rely heavily on a construction of the purpose of the technology. Novel, ill-defined, and morally challenging technologies like NIPT can pose a substantial challenge to policy decision-makers, because the technology’s purpose may be conceptualized in different, sometimes contradictory ways (Giacomini et al., 2013).

In the case of NIPT, ethical and economic analyses suggest conflicting purposes. Ethical and social analyses usually suggest that NIPT’s purpose should be to facilitate informed choice and reproductive autonomy (Deans & Newson, 2012; Dondorp & Lith, 2015; Jong & Wert, 2015). In contrast to this approach, cost-effectiveness analyses (CEAs) of NIPT tend to use outcome measures such as cost per additional chromosomal abnormality detected, cost per additional termination, and cost savings per disabled child not born. CEAs measuring health related quality of life (QALYs) usually measure only maternal QALYs, and include no loss of QALYs for fetuses that might have become babies with a genetic condition (Goldhaber-Fiebert & Brandeau, 2015). These outcome measures might have the normative effect of framing NIPT as a project that becomes more ‘cost effective’ (or worthwhile) only when it prevents a sufficient number of births affected by genetic disability. Members of the disability community have raised serious ethical concerns about framing the purpose of prenatal testing technology in this way.
(Jong & Wert, 2015; Mahowald, 2007; Parens & Asch, 2000). Furthermore, the use of these outcome measures puts policy decision-makers in a challenging situation, because one component of an assessment of NIPT—the economic evaluation including CEA—is evaluating NIPT’s ability to deliver on goals that another component—the social or ethical analysis—suggests it should not have.

This tension between clinical outcomes used in CEA or CUA and ethical or social analyses of how a technology should be used is not unique to NIPT. Morally challenging health technologies may often be evaluated for their cost-effectiveness in achieving clinical outcomes that are socially controversial or ethically problematic; for example, see the ethical arguments against framing pediatric cochlear implants as a technology to cure deafness as a ‘disease’ (Giacomini et al., 2013). However, there is no consensus, in method or theory, on ways in which economists might explicitly consider the ethical or social ‘value’ of a morally challenging health technology when choosing outcome measures for health economic assessment.

This discussion of value touches on a wider question, related to what can be included in the ‘evaluative space’ (Sen, 1993) of health economic assessments—in other words, what kind of information should be considered. Briefly and broadly, the classical answer is the ‘welfarist approach’: that the evaluative space in economic assessment should be limited to individual preferences, or ‘utility’. However, much modern work in health economics has moved beyond a narrow focus on individual utility, towards an ‘extra-welfarist’ approach (Meltzer et al., 2016). Extra-welfarism does not limit its evaluative space to individual utility, and may include utility information but may also evaluate other outcomes, like states and characteristics, or weigh

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utilities from different populations differently. Extra-welfarism also considers stakeholders beyond directly affected individuals (Brouwer et al., 2008).

In health economics, extra-welfarist analyses typically focus solely on health outcomes (e.g. number of terminations, QALYs) (Coast et al., 2008b). However, as we have described, these may conflict with ethical assessments of NIPT. Many health policy decision-making organizations have explicitly adopted an extra-welfarist approach, or prioritized the inclusion of diverse stakeholder perspectives in their analyses (CADTH, 2017). This provides scope, both within extra-welfarist economic theory and within relevant evaluative bodies, for selecting outcome measures informed by ethical as well as social and individual conceptions of value.

In this paper, we provide a theoretical argument for making capabilities (Anand, 2005a; Nussbaum, 2003; Sen, 1993) the evaluative space for economic assessments of morally challenging technologies such as NIPT. Through qualitative analysis, we demonstrate that NIPT can be conceptualized and evaluated as a technology that affects the size, value, and richness of one’s capability set. In doing so, we provide an example of how the capabilities approach may contribute to economic assessments of NIPT that can accommodate ethical and social perspectives on how these technologies ought to be used.

2. The Capabilities Approach and NIPT

The capabilities approach was developed by Amartya Sen as an alternative to standard utilitarian welfare economics (Sen, 1993), and was highly influential in the development of extra-welfarist approaches in health economics (Brouwer et al., 2008). Its central normative proposition is that wellbeing assessments should be based on “what people can do” (their
capabilities) as opposed to “what they actually do” (their functionings) (Anand, 2005a p. 299).

In practical terms, a person’s ‘capability set’ is the set of things they realistically have the opportunity to achieve (Anand, 2005a; Sen, 1993; Sen, 2001).

Sen and other capability theorists have argued that welfarist economics is limited in its ability to assess wellbeing because it takes as its informational basis the amount of benefit or pleasure (utility) that individuals derive from certain functionings, and because this utility must necessarily be measured through expressed preferences (Nussbaum, 2003; Sen, 2001). Both of these aspects are subject to critique.

First, the amount of utility a person derives from a particular functioning is not necessarily well representative of wellbeing. Consider a person who is not eating because they cannot afford food and a person who is not eating because they are fasting. At the level of functioning, the two people are equally well off. But a capabilities analysis would suggest their levels of wellbeing are quite different—the latter was capable of eating, and so may be better off than the former, who had no choice in the matter (Sen, 1993).

Second, preferences are adaptive—they adjust to inequalities and oppressions. People may not express the desire for certain functionings because they have been conditioned not to expect them, and adjust their desires to what they see as possible. In these cases, a focus on preferences alone may obscure significant disadvantage or deprivation (Sen, 2001).

The capabilities approach has already been used to create evaluation metrics in a number of health contexts, including public health (Lorgelly et al., 2015), older populations (Coast et al., 2008a), adult health, (Al-Janabi et al., 2012), mental health (Simon et al., 2013), women’s health (Greco et al., 2015), chronic pain (Kinghorn et al., 2015), and end-of-life care (Huynh et al.,
In these domains, the capabilities approach has provided a substantive theoretical and ethical alternative to welfare-based economic assessments which focus only on utility; one which recognizes that a person’s direct health outcomes may not represent their overall wellbeing (Coast et al., 2008a). It this section, we discuss how a capabilities approach might be considered in the context of NIPT.

A capabilities approach suggests ways in which people might value NIPT that go beyond clinical outcomes or quality of life. For example, the capabilities approach might ask how the availability or routinization of NIPT changes the valuable options available to prospective parents and families. Does NIPT enhance one’s ability to make an informed choice about terminating a pregnancy, or obtain necessary intrapartum and neonatal care at a tertiary hospital? Qualitative studies of women’s and physician’s experiences with NIPT suggest these types of outcomes are valued, but there is a significant gap between this research and cost effectiveness analyses which measure only health outcomes (Lewis et al., 2013; Tiller et al., 2015; Vanstone et al., 2015a).

In addition to asking which options NIPT enhances, a capabilities approach may encourage economists and decision makers to ask which capabilities are diminished by NIPT. For example, a woman might value the capability to make the informed choice not to undergo NIPT, or the capability to raise a disabled child if she wants to; both of which might be lessened in an environment where NIPT is routinized. This focus on options not chosen or opportunities not available reflects a consideration of adaptive preferences, and is a strong deviation from welfarist economics, which assumes that a person’s expressed preferences are the best indication of their values.
This consideration of adaptive preferences relates well to the ethics literature on prenatal testing. Scholars from diverse fields, including feminist and disability theorists, have long emphasized that the choice to undergo prenatal testing is informed by social context. Discourse around prenatal testing is influenced by the clinical and socio-cultural assumption that disability itself is responsible for the reduced life chances of people with disabilities (Albrecht & Devlieger, 1999; Parens & Asch, 2000), and the choice to test is made in the context of medical information systems with complex bearings on reproductive autonomy. Clinicians and counselors may provide limited or selective information about disability and NIPT (Leach, 2015); routinization of testing may erode opportunities for informed choice (Jong & Wert, 2015); and there may be pressure to test, with women being judged irrational or irresponsible for refusing a test that carries no risk of miscarriage (Lewis & Chitty, 2015).

Of course, assessing which capabilities might be enhanced or diminished by NIPT raises its own challenges. Unlike welfarist economics, which bases value on expressed choices and desires, the capabilities approach must explicitly select which capabilities are relevant to wellbeing in a particular context (Sen, 1993). This selection of capabilities for evaluation might be considered paternalistic, and also presents a challenge in a pluralistic society where people will prioritize different capabilities (Carter, 2014).

Approaches to selecting capabilities for assessment

Capability wellbeing assessments developed specifically for use in health contexts have taken a variety of approaches to developing lists of valuable capabilities. One approach is to develop capabilities based on the relevant populations’ expressed views on wellbeing (Al-Janabi...
et al., 2012; Grewal, 2006). Another approach uses empirical capabilities lists. Philosopher Martha Nussbaum developed a list of ten Central Human Capabilities, which includes elements such as Life, Bodily Health, and Practical Reason (Nussbaum, 2003). Anand argues that Nussbaum’s capabilities list could work as a “checklist indicating which dimensions one may want to include in analysis”, though the particular dimensions and specifications would depend on the context (Anand, 2005b p. 1283).

Nussbaum’s list has been criticized for its claims to universality (Jaggar, 2006). However, researchers have developed methods for adapting Nussbaum’s ten Central Human Capabilities to specific contexts, including qualitative and quantitative solicitation of stakeholder views (Lorgelly et al., 2015; Simon et al., 2013). We used Nussbaum’s list as a starting point to analyze data from women who have personal experience with NIPT in order to expand and refine the capabilities relevant to this technology.

3. Methods

Study Data

We conducted a secondary analysis of qualitative data previously collected to investigate women’s values, experiences and opinions related to NIPT (Vanstone et al. 2015a). A comprehensive description of methods is available in the original paper. The data were collected from semi-structured in-depth interviews with 38 women in Ontario, Canada who had personal experience with NIPT.
Data collection took place between April and November 2014. At this time, NIPT had only recently begun to be covered by public health insurance for women with high-risk indications in Ontario. Interview participants were first recruited from a high-risk prenatal diagnostic unit at a tertiary hospital using convenience sampling techniques. After initial analysis, we used online ads on pregnancy websites, snowball sampling techniques, and recruitment posters in private blood labs to purposively select participants with more diverse NIPT experiences, including women who were not eligible for public funding and those who had declined NIPT. This study received research ethics approval from the Hamilton Integrated Research Ethics Board (Review number 14-056).

Data Analysis

We used a constructivist orientation to directed qualitative content analysis (DQCA) to analyze the interviews. DQCA is primarily descriptive, and oriented towards summarizing textual data, respecting context and latent content, using categories from established theory (Drisko & Maschi, 2015).

For this analysis, we operationalized a ‘capability’ as something that a woman could do or achieve as a result of NIPT. The concept of a capability aligned with the concepts of opportunity or option. However, participants were not asked about capabilities directly. That is, the capabilities approach was neither explained during the interview nor used in developing interview questions.

Data were analyzed using line by line coding techniques (Charmaz, 2014), expanding and modifying ten preliminary codes generated deductively from Nussbaum’s ten Central Human...
Capabilities. After a first round of coding, inductive analysis was used to consider the data that suggested new aspects of Nussbaum’s capabilities, or which fit the definition of a capability but were not adequately described by Nussbaum’s list. We then progressed to focused coding around refined concepts of capabilities (Charmaz, 2014).

Initial coding was conducted independently by two researchers (MK and AC). The remainder of the coding was conducted by researcher MK, with frequent consultation with MV to discuss emergent findings, and reflective memos were kept by MK and reviewed by MV in order to facilitate reflexive engagement. NVivo 11 software was used for data management. Coding continued until data saturation had been achieved. We determined saturation through discussion, and the coding of two more interviews (27 participants).

4. Results

These results describe capabilities related to NIPT as identified in interviews with 27 women, whose characteristics are described in Table 1. We used all 10 of Nussbaum’s characteristics as the initial basis for codes, and found data that fit eight (Life; Bodily Health; Bodily Integrity; Senses, Imagination, and Thought; Emotions; Practical Reason; Affiliation). We did not find data to support two (Play, Other Species). We also identified one additional capability not included in Nussbaum’s list: Care-Taking. The capabilities overlap in different dimensions—for example, the ability to protect oneself from an unwanted miscarriage as a result of invasive prenatal genetic testing was categorized as a component of the Bodily Health capability, but is also linked to Practical Reason, because women valued this ability differently depending on their reproductive goals.
In this section, we describe how the nine capabilities we identified in the data relate to NIPT. Although the Play and Other Species capabilities were not identified in this dataset, their absence does not necessarily mean they are irrelevant to an assessment of how NIPT affects women’s capabilities. Rather, it likely reflects the secondary nature of our analysis—we were not able to ask specific questions in the interviews, and topics related to these capabilities did not come up spontaneously in the discussion.

Life

Women conceptualized this capability as most relevant not to themselves but to their potential children, as the ability to find out if the fetus they were carrying had a genetic disorder that would severely impact their quality of life,

I think when you’re talking about genetic screening, you’re talking about things that are gonna severely impact their quality of life…

Many women also thought that NIPT should be reserved for cases where the capability of life might genuinely be threatened by a genetic disability, and not be used to detect nonlife-threatening or -altering conditions, “Yeah, I think there’s got to be a limit, but I think, certainly, anything that could really impact on the child’s quality of life, in a significant way”

Bodily Health

In the context of NIPT, discussions corresponding to this capability related overwhelmingly to reproductive health. Women conceptualized NIPT as a technology that might improve their reproductive health, primarily by helping them avoid iatrogenic miscarriage from an invasive prenatal genetic test like amniocentesis:
“If I had to pay for it, I would borrow from my friends or relatives. But I would just do anything possible to avoid a miscarriage.”

NIPT’s perceived ability to help women avoid iatrogenic miscarriage was mediated by its accuracy, which women perceived as high enough to make NIPT a good alternative to an invasive test, “It just seemed like a great alternative that might give us the same clarity as doing an amniocentesis without the risk”

**Bodily Integrity**

In the context of NIPT, the bodily integrity capability was related to women’s ability to have choice in matters of reproduction, specifically prenatal care. It was very closely related to the capability of ‘Control Over One’s Environment’, which in this context was the prenatal healthcare system. Women explained that they valued knowing being told about NIPT because it made them feel empowered in making decisions about their reproductive care, “I just really think that women should be given ownership of the information and they can decide what they want to do”.

**Senses, Imagination, and Thought**

This capability was linked to the ability to deliberate, seek information about different tests, and consider one’s personal views on pregnancy and disability in a way that was cultivated by fully understanding one’s prenatal genetic testing options. This capability was about more than just knowing about NIPT—it was about being able to attain a good enough understanding to really consider what the test and results might mean, “I mean, even just soul searching yourself and knowing, okay so what am I going to do with these results regardless?”
A lack of education about NIPT impeded women’s ability to reflect on its implications,

“Well I think a lot of women don’t really want to think about it but I think they need
to understand what the possible outcomes are. Like I think everybody knows that
Down’s syndrome is trisomy 21 but there seems to be a lot of vagueness and even a
lot of confusion about the testing itself and about what they’re actually looking for.”

As did a lack of adequate counselling and support, “I just wanted to talk to somebody
about what, if I do continue with the pregnancy, what that means, like, what I'm going to be
dealing with. Nobody there could really tell me anything”

Emotions

Fear, stress, and anxiety were recurrent themes in women’s discussions of NIPT,
whether they ultimately decided to have the test or not. Multiple women explained that
they wanted NIPT because it could give them some “peace of mind” or “relief.” We
identified peace of mind and reassurance as functionings that many women found
desirable, and their decision to have or not have NIPT was related to how they felt it might
contribute to their achieving this functioning. Both women who accepted and declined
NIPT explained this decision in terms of managing their stress and anxiety. This woman
describes how declining NIPT contributed to lowering her stress:

“I was going to go for the blood work but then I just was like, like you know what, I
would rather not think about it, I would rather not stress about it.”

Other women described that participating in NIPT could lower stress because it
made it possible to get further information without having to undergo invasive testing:

“if there was no NIPT, I'd be left with a positive result from my Integrated Prenatal
Screening, not willing to do an amniocentesis because I'm not willing to you know,

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go with that risk, so the rest of my pregnancy I have that added stress of thinking that I have a high high chance of having you know, a baby with Down Syndrome”

Practical Reason

Practical reason was conceptualized as the ability to engage in critical reflection and planning in order to make what one felt were ‘good’ or ‘the best possible’ decisions about one’s life. NIPT enhanced this capability because it was conceptualized as a technology that helped women “make the right decisions and make the decisions that are best for their family.” This was one of the most prominent capabilities in our data, and one of the most complex.

Two elements were central to the way NIPT enhanced women’s abilities to engage in planning and make ‘good’ decisions about prenatal testing: the accuracy of NIPT, and the timing of NIPT. NIPT’s accuracy made women feel like they could trust the test results, and even women who did not have NIPT sometimes thought of it as a ‘backup’ for tests with a higher false-positive rate, “So that kind of helped me make my decision to say, okay, well, if I get a negative, then I’m not going to be concerned, but if I get a positive, I’ll just go and take the NIPT”.

Having NIPT at different times was perceived to open up different sets of options with regards to pregnancy management and termination:

“If you have a positive result, if you get a result at 10 or 11, well let's say 11 weeks or 12 weeks, your decision about what you do with that information could be different if you get it later in the pregnancy.”

However, it was not entirely clear that women always knew from the outset how the information they got from NIPT would affect their decisions about their pregnancy. Women
often framed NIPT as a technology that could give them the capability to make informed decisions about pregnancy, without explicitly specifying a predetermined goal:

“No we didn't discuss, we still haven't discussed what we would do with that information once we got it but we knew we wanted to go forward to get the information.”

Practical Reason was also linked to wide range of interventions beyond NIPT, including diagnostic prenatal genetic testing, termination, social services for children with disabilities and their families. So, NIPT cannot be said to facilitate women’s Practical Reasoning abilities in isolation, but rather in conjunction with other technologies.

Affiliation

The affiliation capability was conceptualized as the ability to live in social communities where all members are valued and supported. Much of the discussion relevant to this capability related to concerns that providers’ descriptions of NIPT did not address the lives, role, and place of disabled people in society,

“It talks to you all about your risks of getting it and what it is, but there is nothing in here about, you know, how you deal with it. So, in some ways it’s unfortunate because, [pause, sigh] I don’t know. It gets to a bigger societal question, right, about acceptance of people and their place in society”

Women also discussed NIPT’s potential to constrain Affiliation, and emphasized the importance of continued support for families and children with disabilities, “they still need the support from the government and the society to help make the process of raising this child more positive”, and brought up concerns that, down the road, NIPT could be used to discriminate based on cosmetic traits, “Are they going to start testing for, I want a kid that’s going to be the tallest? I want
Control Over One’s Environment

This capability was closely related to the Practical Reason and Bodily Integrity capabilities. Control Over One’s Environment is distinct in that it deals specifically with the ability to participate in choices that govern what is available in the larger medical or prenatal care system. To some degree, both the Practical Reason and Bodily Integrity capabilities are dependent on Control Over One’s Environment, because one’s ability to plan and think (Practical Reason) and choice in individual matters of reproduction (Bodily Integrity) are constrained when valued options aren’t represented in the medical care system. In this context, discussions centered around women’s strong views that the healthcare mandate should be to tell all pregnant women about NIPT, not just high risk women eligible for public coverage “I think people should be offered. It’s their choice if they want to do it or not, or pay it or not. But as long as the science is there, I think that people should be made aware of it.”

A new capability: Care-Taking (for existing or potential children and family)

The ability to adequately take care of dependents, including children, is not included on Nussbaum’s list of ten Central Human Capabilities. However, we identified in our data that caring for the anticipated “child” is a powerful motivator for the decision to get NIPT. The data that supported this new capability centered around the magnitude and type of care-taking inherent in having a child, with or without disabilities. It was identified as a distinct ‘Care-
Taking’ capability because ability to perform this care-taking was not mutually inclusive or exclusive of other related capabilities, like Emotion or Affiliation. For example, we found that a person could still exercise all elements of the Emotions capability (obtaining peace of mind, have attachments to others, love their existing and potential children, including those that might have disabilities) without feeling they could adequately take care of existing children or a child with a disability. For example, one woman discussed what would have happened if she hadn’t accessed NIPT:

“I was concerned, largely, with the impact that it would have on my existing son, and also, just concerned about what our child with trisomy 21 would have to go through in his life …I would have a son right now with trisomy 21, which, I mean, if I had him I would love him, in some respect it would be great, but I don’t know how terrible his disability would be, because you can't know that, but it could be pretty awful.”

In a way that reflects how she valued NIPT not necessarily because it enhanced her capability to love or feel emotions toward her potential and existing children (she would have done that without NIPT), but because enhanced her practical ability to take care of them.

Data related to this capability involved NIPT’s effect on women’s ability to consider or plan to parent a child with special needs: “it’s just more for the parents to prepare for the kids. Like how do they take care of the kids?”, and take care of existing children “my husband grew up with a mentally challenged sister, and it’s not something we wanted for our son.”

5. Discussion

We found that the language and concept of capabilities was highly relevant to women’s experiences and values related to NIPT. Eight of Nussbaum’s ten central capabilities and one newly identified capability relate to options, choices, or states that NIPT gives women the ability
to achieve. Overall, women value NIPT in part because it enlarges their capability set by giving them more and different choices in their prenatal care pathway.

The dimensions and aspects of capabilities we identified in this study will not be universally relevant. The women in our sample were older than the Canadian average (Statistics Canada, 2017), almost all had partners, almost half had a graduate or professional degree, and most lived in cities. Though the individual women in our sample came from diverse backgrounds, they may have had relatively high socioeconomic status overall, which likely affected their conception of capabilities, in particular capabilities that would have been affected by the cost of NIPT. Furthermore, most of the women in our sample were recruited from a high-risk prenatal diagnostic unit at a tertiary hospital, which also may have shaped their ideas of capabilities NIPT could offer, particularly their perception that NIPT could offer emotional reassurance. Although further work with different populations and stakeholder groups will be necessary to develop a list of capabilities to be used in assessments, this analysis demonstrates that NIPT can indeed be conceptualized and evaluated as a technology that affects the size, value, and richness of one’s capability set.

This research has a number of implications for future work in the field of economic evaluations of NIPT. First and foremost, it suggests that capability measures may be used to help economists move beyond ethically problematic assessments of NIPT that rely on clinical and QALY-based outcome measures. This work could form the preliminary basis of an ‘index of capability’ for NIPT, similar to indices of capability for other populations (Al-Janabi et al., 2008; Al-Janabi et al., 2012; Coast et al., 2008a; Huynh et al., 2017; Kinghorn et al., 2015; Lorgelly et al., 2015; Simon et al., 2013). Notably, all of these indices with the exception of the ICECAP-
SCM and OCAP-18 (Huynh et al., 2017; Lorgelly et al., 2015) develop outcome measures for populations, whereas this research was based around a specific intervention. This intervention-specific focus reflects the organic development of this research; our team noticed an important tension in the economic evaluation of NIPT, and set out to explore potential solutions. The next step in this research is to develop our findings into a useable outcome measurement. This will require a decision about whether this measure should be generalized to a population (e.g. an index of capability for pregnant people) or a class of interventions (e.g. an index of capability for people undergoing genetic testing, or prenatal genetic testing).

We are not the first to point out that many health-related outcome measures don’t accurately or adequately capture the value of genetic testing, and prenatal genetic testing in particular. For example, willingness to pay estimates for prenatal genetic screening are not highly sensitive to whether a woman plans to terminate an affected pregnancy or not (Grosse et al., 2008). Patients identify attributes like decision making, satisfaction, and perceived control as benefits of genetic testing (Payne et al., 2007). However, research that translates these findings into economic evaluations of prenatal genetic testing is limited. Direct approaches to eliciting preferences for health states, non-health outcomes, and process attributes related to prenatal genetic testing, (Bishop et al., 2004; Feeny & Tomkins, 2004; Hall et al., 2006) have given us insight into preferences for outcomes associated with prenatal genetic testing, and how prenatal genetic tests should be delivered, but cannot speak to how these technologies affect a larger scope of wellbeing. Furthermore, preference elicitation studies typically do not include stakeholders beyond those directly affected by the technology, including people living with disabilities and larger communities, when developing options or eliciting preferences (Feeny &
Tomkins, 2004). A capabilities approach, however, does not preclude preference elicitation—a capabilities analysis could inform options in discrete choice experiments (Coast et al., 2012).

There has been some discussion of the ‘value of information’ as a construct that could be used to capture the ways in which people value prenatal genetic testing beyond narrow health outcomes (Feeny et al., 2002; Grosse et al., 2008; Ryan et al., 2003). Indeed, it is evident both from our analysis and from many others (Lewis et al., 2013; Tiller et al., 2015) that women do value knowing about NIPT and being able to make informed choices about prenatal genetic testing. However, we would argue that the information itself is only valuable insofar as what it is perceived to provide—in many cases, a sense of reassurance, control, or ’expert’ opinion (Lewis & Chitty, 2015). The capabilities approach directly investigates the ways in which knowledge generated by NIPT is valuable by focusing on the options or choices it does (or does not) create. A capabilities approach could also make it possible to ask important questions about whether prenatal genetic testing technologies are an appropriate or optimal way of achieving certain objectives. For example—prenatal genetic testing may offer ‘emotional reassurance’ to pregnant women, but may also contribute to the medicalized anxiety that necessitates that reassurance (Lippman, 1991).

A capabilities approach to evaluating NIPT does have limitations. In any qualitative development of capability attributes, adaptive values and preferences will influence conceptions of relevant capabilities. In our data, this was most notable in women’s discussions of the Life, Emotion, Practical Reason, and Care-Taking capabilities, which were potentially heavily influenced by socio-cultural perceptions of disability. In all of these areas, many women discussed the idea that NIPT could tell them if the “baby” was “healthy”. This idea, that negative
results from prenatal genetic mean a ‘healthy’ baby, has been discussed extensively in the social, ethical, and disabilities literature on prenatal testing, which points out that disabled people may be perfectly ‘healthy’ (Lippman, 1991; Rapp, 2004). The Care-Taking and Life capabilities in particular may have been influenced by the perception that taking care of a child with a genetic disability is extremely onerous and potentially harmful to existing children; increased availability of social supports to people with disabilities and their families may affect women’s perceptions of their relevance to NIPT.

Our definition of Care-Taking as a new capability also raises interesting questions. Ingrid Robeyns, in her list of capabilities developed to assess gender equity, includes a similar capability (Domestic work and non-market care), and points out that it is different from many capabilities in that engaging in care-taking is not always beneficial to the care-taker (Robeyns, 2003). Thus, even though Care-Taking came out strongly in the data, it’s not clear that it should be included in a list of capabilities relevant to NIPT. Elicited values alone are not a sufficient basis for the development of a capability wellbeing measure, and it will be necessary to involve ethicists, disability activists, and other scholars in the development of an equitable, ethical, capability measure for NIPT and other prenatal genetic tests.

Strengths and Limitations:

To our knowledge, this study is the first to apply the capabilities approach to a prenatal genetic testing technology. However, the data for this study was drawn from a primary study on women’s experiences and values with NIPT. As such, we did not incorporate the language and
concept of capabilities while developing interview questions, and could not ask probing questions or use interviews to refine concepts.

The women in this sample were older and more educated than the average pregnant woman in our population, which may have affected the capabilities they associated with NIPT.

Furthermore, the participants in this study lived in Ontario, Canada and the findings are rooted in the healthcare, policy, and social context of that province.

Conclusion:

This qualitative study explored whether the capabilities approach could be used to assess the value of NIPT in a way that is useful to economists, while also accommodating a wider scope of value suggested by ethical and social analyses. Women identified NIPT as a technology that enhanced their capability set. Eight of Nussbaum’s ten capabilities were relevant to NIPT; all except Play and Other Species. We identified one new capability not on Nussbaum’s list, Care-Taking. A capabilities approach can provide economists and policy decision makers with tools to analyze NIPT based on its potential to promote informed choice, instead of health outcomes of ambiguous social and ethical value. This research adds to extra-welfarist approaches in health economic evaluation, and may help resolve conflicts between economic and ethical evaluations of morally complex health technologies.
References


Table 1 – Characteristics of the study participants

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>N/Median(Range)</th>
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<td>Number of participants</td>
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<tr>
<td>Age</td>
<td>36 (25-45)</td>
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<tr>
<td>Number of existing children</td>
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<tr>
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<td>7</td>
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<tr>
<td>One</td>
<td>13</td>
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<td>Two or Three</td>
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<td>Previous miscarriage or neonatal death*</td>
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<tr>
<td>Second Trimester</td>
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</table>

* This information was volunteered in the interview, more women may have had this experience and chosen not to share