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1 Title: Reconciling ethical and economic conceptions of value in health policy using the
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3

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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
70 ambiguous social and ethical value. More broadly, the capabilities approach may serve to resolve
71 contradictions between ethical and economic evaluations of health technologies, and contribute
72 to extra-welfarist approaches in the assessment of morally complex health technologies.

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4. Capabilities Approach

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6. Prenatal genetic testing

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7. Ethics

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8. Health Policy

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91 **1. Introduction**

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

104

105 *Non-invasive prenatal testing*

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

113 but it is likely that NIPT will eventually be expanded to include a variety of genetic conditions
114 (Hui & Bianchi, 2017).

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

121 NIPT has been rapidly and broadly adopted for prenatal genetic testing, as both a first-tier
122 screening test for disability and as a second-tier screen to reduce the risk of iatrogenic
123 miscarriage from invasive diagnostic procedures (Minear et al., 2015). This expansion has been
124 facilitated by industry imperatives. NIPT technologies were developed by a number of different
125 private companies, and in many places remain private-pay technologies, although some
126 jurisdictions have recently offered coverage through public and private insurers (Minear et al.,
127 2015; Vanstone et al., 2015b).

128 The commercial proliferation of NIPT preceded careful policy decision-making about its
129 use and value (Vanstone et al., 2014). Evidence around NIPT's clinical utility for different
130 conditions and patient populations is still developing, and it is not yet clear how NIPT will
131 integrate with existing prenatal testing technologies (Murdoch et al., 2017). The rapid expansion
132 of NIPT has raised ethical concerns about the routinization of testing and erosion of informed
133 decision-making processes (Deans & Newson, 2012; Lewis et al., 2013). Like other prenatal
134 testing technologies, NIPT is challenged by ambiguously defined social and medical purposes

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135 (Mahowald, 2007). Furthermore, ethical and policy analyses of NIPT are complicated because
136 NIPT is not a homogeneous entity; it can refer to a number of slightly different methods for
137 analyzing fetal genetic material, and is used by people and practitioners in different ways and for
138 different purposes (Vanstone et al., 2015b).

139

140 *Economic and Ethical Evaluations of NIPT*

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

145 In the case of NIPT, ethical and economic analyses suggest conflicting purposes. Ethical
146 and social analyses usually suggest that NIPT's purpose should be to facilitate informed choice
147 and reproductive autonomy (Deans & Newson, 2012; Dondorp & Lith, 2015; Jong & Wert,
148 2015). In contrast to this approach, cost-effectiveness analyses (CEAs) of NIPT tend to use
149 outcome measures such as cost per additional chromosomal abnormality detected, cost per
150 additional termination, and cost savings per disabled child not born. CEAs measuring health
151 related quality of life (QALYs) usually measure only maternal QALYs, and include no loss of
152 QALYs for fetuses that might have become babies with a genetic condition (Goldhaber-Fiebert
153 & Brandeau, 2015). These outcome measures might have the normative effect of framing NIPT
154 as a project that becomes more 'cost effective' (or worthwhile) only when it prevents a sufficient
155 number of births affected by genetic disability. Members of the disability community have raised
156 serious ethical concerns about framing the purpose of prenatal testing technology in this way

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157 (Jong & Wert, 2015; Mahowald, 2007; Parens & Asch, 2000). Furthermore, the use of these
158 outcome measures puts policy decision-makers in a challenging situation, because one
159 component of an assessment of NIPT—the economic evaluation including CEA—is evaluating
160 NIPT's ability to deliver on goals that another component—the social or ethical analysis—
161 suggests it should not have.

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

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

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

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

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

195

196 **2. The Capabilities Approach and NIPT**

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

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201 capabilities) as opposed to “what they actually do” (their functionings) (Anand, 2005a p. 299) .

202 In practical terms, a person’s ‘capability set’ is the set of things they realistically have the

203 opportunity to achieve (Anand, 2005a; Sen, 1993; Sen, 2001).

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

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

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

219 The capabilities approach has already been used to create evaluation metrics in a number
220 of health contexts, including public health (Lorgelly et al., 2015), older populations (Coast et al.,
221 2008a), adult health, (Al-Janabi et al., 2012), mental health (Simon et al., 2013), women’s health
222 (Greco et al., 2015), chronic pain (Kinghorn et al., 2015), and end-of-life care (Huynh et al.,

223 2017). In these domains, the capabilities approach has provided a substantive theoretical and
224 ethical alternative to welfare-based economic assessments which focus only on utility; one which
225 recognizes that a person's direct health outcomes may not represent their overall wellbeing (Coast
226 et al., 2008a). In this section, we discuss how a capabilities approach might be considered in the
227 context of NIPT.

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

237 In addition to asking which options NIPT enhances, a capabilities approach may
238 encourage economists and decision makers to ask which capabilities are *diminished* by NIPT.
239 For example, a woman might value the capability to make the informed choice not to undergo
240 NIPT, or the capability to raise a disabled child if she wants to; both of which might be lessened
241 in an environment where NIPT is routinized. This focus on options *not* chosen or opportunities
242 *not* available reflects a consideration of adaptive preferences, and is a strong deviation from
243 welfarist economics, which assumes that a person's expressed preferences are the best indication
244 of their values.

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245 This consideration of adaptive preferences relates well to the ethics literature on prenatal
246 testing. Scholars from diverse fields, including feminist and disability theorists, have long
247 emphasized that the choice to undergo prenatal testing is informed by social context. Discourse
248 around prenatal testing is influenced by the clinical and socio-cultural assumption that disability
249 itself is responsible for the reduced *life chances* of people with disabilities (Albrecht &
250 Devlieger, 1999; Parens & Asch, 2000), and the choice to test is made in the context of medical
251 information systems with complex bearings on reproductive autonomy. Clinicians and
252 counselors may provide limited or selective information about disability and NIPT (Leach,
253 2015); routinization of testing may erode opportunities for informed choice (Jong & Wert, 2015);
254 and there may be pressure to test, with women being judged irrational or irresponsible for
255 refusing a test that carries no risk of miscarriage (Lewis & Chitty, 2015).

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

262

263 *Approaches to selecting capabilities for assessment*

264 Capability wellbeing assessments developed specifically for use in health contexts have
265 taken a variety of approaches to developing lists of valuable capabilities. One approach is to
266 develop capabilities based on the relevant populations' expressed views on wellbeing (Al-Janabi

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267 et al., 2012; Grewal, 2006). Another approach uses empirical capabilities lists. Philosopher
268 Martha Nussbaum developed a list of ten Central Human Capabilities, which includes elements
269 such as Life, Bodily Health, and Practical Reason (Nussbaum, 2003). Anand argues that
270 Nussbaum's capabilities list could work as a "checklist indicating which dimensions one may
271 want to include in analysis", though the particular dimensions and specifications would depend
272 on the context (Anand, 2005b p. 1283).

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

279

280 **3. Methods**

281 *Study Data*

282 We conducted a secondary analysis of qualitative data previously collected to investigate
283 women's values, experiences and opinions related to NIPT (Vanstone et. al 2015a). A
284 comprehensive description of methods is available in the original paper. The data were collected
285 from semi-structured in-depth interviews with 38 women in Ontario, Canada who had personal
286 experience with NIPT.

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288 Data collection took place between April and November 2014. At this time, NIPT had
289 only recently begun to be covered by public health insurance for women with high-risk
290 indications in Ontario. Interview participants were first recruited from a high-risk prenatal
291 diagnostic unit at a tertiary hospital using convenience sampling techniques. After initial
292 analysis, we used online ads on pregnancy websites, snowball sampling techniques, and
293 recruitment posters in private blood labs to purposively select participants with more diverse
294 NIPT experiences, including women who were not eligible for public funding and those who had
295 declined NIPT. This study received research ethics approval from the Hamilton Integrated
296 Research Ethics Board (Review number 14-056).

297

298 *Data Analysis*

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

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

308 Data were analyzed using line by line coding techniques (Charmaz, 2014), expanding and
309 modifying ten preliminary codes generated deductively from Nussbaum's ten Central Human

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310 Capabilities. After a first round of coding, inductive analysis was used to consider the data that
311 suggested new aspects of Nussbaum's capabilities, or which fit the definition of a capability but
312 were not adequately described by Nussbaum's list. We then progressed to focused coding around
313 refined concepts of capabilities (Charmaz, 2014).

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

320

321 **4. Results**

322 These results describe capabilities related to NIPT as identified in interviews with 27
323 women, whose characteristics are described in Table 1. We used all 10 of Nussbaum's
324 characteristics as the initial basis for codes, and found data that fit eight (Life; Bodily Health;
325 Bodily Integrity; Senses, Imagination, and Thought; Emotions; Practical Reason; Affiliation).
326 We did not find data to support two (Play, Other Species). We also identified one additional
327 capability not included in Nussbaum's list: Care-Taking. The capabilities overlap in different
328 dimensions—for example, the ability to protect oneself from an unwanted miscarriage as a result
329 of invasive prenatal genetic testing was categorized as a component of the Bodily Health
330 capability, but is also linked to Practical Reason, because women valued this ability differently
331 depending on their reproductive goals.

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332 In this section, we describe how the nine capabilities we identified in the data relate to
333 NIPT. Although the Play and Other Species capabilities were not identified in this dataset, their
334 absence does not necessarily mean they are irrelevant to an assessment of how NIPT affects
335 women's capabilities. Rather, it likely reflects the secondary nature of our analysis—we were not
336 able to ask specific questions in the interviews, and topics related to these capabilities did not
337 come up spontaneously in the discussion.

338

339 *Life*

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

343 I think when you're talking about genetic screening, you're talking about things that
344 are gonna severely impact their quality of life...

345

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

350

351 *Bodily Health*

352 In the context of NIPT, discussions corresponding to this capability related
353 overwhelmingly to reproductive health. Women conceptualized NIPT as a technology that might
354 improve their reproductive health, primarily by helping them avoid iatrogenic miscarriage from
355 an invasive prenatal genetic test like amniocentesis:

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356 “If I had to pay for it, I would borrow from my friends or relatives. But I would just
357 do anything possible to avoid a miscarriage.”

358
359 NIPT’s perceived ability to help women avoid iatrogenic miscarriage was mediated by its
360 accuracy, which women perceived as high enough to make NIPT a good alternative to an
361 invasive test, “It just seemed like a great alternative that might give us the same clarity as doing
362 an amniocentesis without the risk”

363
364 *Bodily Integrity*

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

372
373 *Senses, Imagination, and Thought*

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

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380
381 A lack of education about NIPT impeded women's ability to reflect on its implications,

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

386
387 As did a lack of adequate counselling and support, "I just wanted to talk to somebody
388 about what, if I do continue with the pregnancy, what that means, like, what I'm going to be
389 dealing with. Nobody there could really tell me anything"

390

391 *Emotions*

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

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

402

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

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

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407 go with that risk, so the rest of my pregnancy I have that added stress of thinking that
408 I have a high high chance of having you know, a baby with Down Syndrome”
409

410 *Practical Reason*

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

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

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

425 “If you have a positive result, if you get a result at 10 or 11, well let's say 11 weeks
426 or 12 weeks, your decision about what you do with that information could be
427 different if you get it later in the pregnancy.”
428

429 However, it was not entirely clear that women always knew from the outset how the
430 information they got from NIPT would affect their decisions about their pregnancy. Women

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431 often framed NIPT as a technology that could give them the capability to make informed
432 decisions about pregnancy, without explicitly specifying a predetermined goal:

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

436
437 Practical Reason was also linked to wide range of interventions beyond NIPT, including
438 diagnostic prenatal genetic testing, termination, social services for children with disabilities and
439 their families. So, NIPT cannot be said to facilitate women's Practical Reasoning abilities in
440 isolation, but rather in conjunction with other technologies.

441

442 *Affiliation*

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

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

451

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

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457 somebody that's pretty, or somebody that's handsome, or somebody that's the smartest. So it's a
458 slippery slope.”

459

460 *Control Over One's Environment*

461 This capability was closely related to the Practical Reason and Bodily Integrity
462 capabilities. Control Over One's Environment is distinct in that it deals specifically with the
463 ability to participate in choices that govern what is available in the larger medical or prenatal
464 care system. To some degree, both the Practical Reason and Bodily Integrity capabilities are
465 dependent on Control Over One's Environment, because one's ability to plan and think (Practical
466 Reason) and choice in individual matters of reproduction (Bodily Integrity) are constrained when
467 valued options aren't represented in the medical care system. In this context, discussions
468 centered around women's strong views that the healthcare mandate should be to tell all pregnant
469 women about NIPT, not just high risk women eligible for public coverage “I think people should
470 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
471 is there, I think that people should be made aware of it.”

472

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

474 The ability to adequately take care of dependents, including children, is not included on
475 Nussbaum's list of ten Central Human Capabilities. However, we identified in our data that
476 caring for the anticipated “child” is a powerful motivator for the decision to get NIPT. The data
477 that supported this new capability centered around the magnitude and type of care-taking
478 inherent in having a child, with or without disabilities. It was identified as a distinct ‘Care-

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479 Taking' capability because ability to perform this care-taking was not mutually inclusive or
480 exclusive of other related capabilities, like Emotion or Affiliation. For example, we found that a
481 person could still exercise all elements of the Emotions capability (obtaining peace of mind, have
482 attachments to others, love their existing and potential children, including those that might have
483 disabilities) without feeling they could adequately take care of existing children or a child with a
484 disability. For example, one women discussed what would have happened if she hadn't accessed
485 NIPT:

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

492
493 In a way that reflects how she valued NIPT not necessarily because it enhanced her capability to
494 love or feel emotions toward her potential and existing children (she would have done that
495 without NIPT), but because enhanced her practical ability to take care of them.

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

500

501 **5. Discussion**

502 We found that the language and concept of capabilities was highly relevant to women's
503 experiences and values related to NIPT. Eight of Nussbaum's ten central capabilities and one
504 newly identified capability relate to options, choices, or states that NIPT gives women the ability

505 to achieve. Overall, women value NIPT in part because it enlarges their capability set by giving
506 them more and different choices in their prenatal care pathway.

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

520 This research has a number of implications for future work in the field of economic
521 evaluations of NIPT. First and foremost, it suggests that capability measures may be used to help
522 economists move beyond ethically problematic assessments of NIPT that rely on clinical and
523 QALY-based outcome measures. This work could form the preliminary basis of an 'index of
524 capability' for NIPT, similar to indices of capability for other populations (Al-Janabi et al., 2008;
525 Al-Janabi et al., 2012; Coast et al., 2008a; Huynh et al., 2017; Kinghorn et al., 2015; Lorgelly et
526 al., 2015; Simon et al., 2013). Notably, all of these indices with the exception of the ICECAP-

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527 SCM and OCAP-18 (Huynh et al., 2017; Lorgelly et al., 2015) develop outcome measures for
528 populations, whereas this research was based around a specific intervention. This intervention-
529 specific focus reflects the organic development of this research; our team noticed an important
530 tension in the economic evaluation of NIPT, and set out to explore potential solutions. The next
531 step in this research is to develop our findings into a useable outcome measurement. This will
532 require a decision about whether this measure should be generalized to a population (e.g. an
533 index of capability for pregnant people) or a class of interventions (e.g. an index of capability for
534 people undergoing genetic testing, or prenatal genetic testing).

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

549 Tomkins, 2004). A capabilities approach, however, does not preclude preference elicitation—a
550 capabilities analysis could inform options in discrete choice experiments (Coast et al., 2012).

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

565 A capabilities approach to evaluating NIPT does have limitations. In any qualitative
566 development of capability attributes, adaptive values and preferences will influence conceptions
567 of relevant capabilities. In our data, this was most notable in women’s discussions of the Life,
568 Emotion, Practical Reason, and Care-Taking capabilities, which were potentially heavily
569 influenced by socio-cultural perceptions of disability. In all of these areas, many women
570 discussed the idea that NIPT could tell them if the “baby” was “healthy”. This idea, that negative

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571 results from prenatal genetic mean a 'healthy' baby, has been discussed extensively in the social,
572 ethical, and disabilities literature on prenatal testing, which points out that disabled people may
573 be perfectly 'healthy' (Lippman, 1991; Rapp, 2004). The Care-Taking and Life capabilities in
574 particular may have been influenced by the perception that taking care of a child with a genetic
575 disability is extremely onerous and potentially harmful to existing children; increased availability
576 of social supports to people with disabilities and their families may affect women's perceptions
577 of their relevance to NIPT.

578 Our definition of Care-Taking as a new capability also raises interesting questions. Ingrid
579 Robeyns, in her list of capabilities developed to assess gender equity, includes a similar
580 capability (Domestic work and non-market care), and points out that it is different from many
581 capabilities in that engaging in care-taking is not always beneficial to the care-taker (Robeyns,
582 2003). Thus, even though Care-Taking came out strongly in the data, it's not clear that it should
583 be included in a list of capabilities relevant to NIPT. Elicited values alone are not a sufficient
584 basis for the development of a capability wellbeing measure, and it will be necessary to involve
585 ethicists, disability activists, and other scholars in the development of an equitable, ethical,
586 capability measure for NIPT and other prenatal genetic tests.

587

588 *Strengths and Limitations:*

589 To our knowledge, this study is the first to apply the capabilities approach to a prenatal
590 genetic testing technology. However, the data for this study was drawn from a primary study on
591 women's experiences and values with NIPT. As such, we did not incorporate the language and

592 concept of capabilities while developing interview questions, and could not ask probing
593 questions or use interviews to refine concepts.

594 The women in this sample were older and more educated than the average pregnant
595 woman in our population, which may have affected the capabilities they associated with NIPT.
596 Furthermore, the participants in this study lived in Ontario, Canada and the findings are rooted in
597 the healthcare, policy, and social context of that province.

598

599 **Conclusion:**

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

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745

Table 1 – Characteristics of the study participants

Characteristic	N/Median(Range)
Number of participants	27
Age	36 (25-45)
Number of existing children	
Zero	7
One	13
Two or Three	7
Partnered at time of experience with NIPT	27
Previous miscarriage or neonatal death*	5
Education	
High school	1
College/undergraduate degree	15
Graduate or professional degree	11
Occupation	
Stay at home mother, homemaker	5
Education	5
Healthcare	3
Business	7
Manufacturing	1
Personal services	1
Government	1
Law	1
Customer service	1
Not disclosed	2
City	16
Town or suburb	9
Rural	2

Testing & coverage	
Eligible for publicly funded NIPT & accepted testing	17
Eligible for publicly funded NIPT & declined testing	4
Ineligible for publicly funded NIPT & accepted testing	4
Ineligible for publicly funded NIPT & declined testing	2
Trimester offered	
Preconception	2
First Trimester	6
Second Trimester	19

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