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Research

Benefits, challenges and ethical principles associated with implementing noninvasive prenatal testing: a Delphi study

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Abstract

Background: Noninvasive prenatal testing is a recent technology that provides some genetic information about the fetus through the analysis of cell-free fetal DNA circulating in maternal blood. We aimed to identify the benefits, challenges and guiding ethical principles most relevant to the clinical integration of noninvasive prenatal testing in Canada, according to experts throughout the country.

Methods: We conducted a 3-round Delphi study involving Canadian experts of contemporary discussions about the ethical and societal implications of prenatal testing and genomic technologies. In round 1, we asked participants to identify clinical benefits and challenges related to the implementation of noninvasive prenatal testing in Canada, and the ethical principles they think should guide it. In round 2, we asked participants to select the most important elements stated by their peers. In round 3, participants were informed of the aggregated results from round 2, and invited to revise or confirm their selection.

Results: Round 1 had a participation rate of 20.2%, and involved 61 participants. Subsequent rounds 2 and 3 had retention rates of 95.1% ($n = 58$) and 84.5% ($n = 49$), respectively. Through these discussions, we identified 3 lists of benefits ($n = 10$), challenges ($n = 27$), and ethical principles ($n = 16$) prioritized by Canadian experts as being most relevant to the implementation of noninvasive prenatal testing in Canada.

Interpretation: Although multiple and diverse potential issues were identified, Canadian experts agreed on 2 sets of requirements for the responsible implementation of noninvasive prenatal testing in Canada. Interdisciplinary appraisals may be instrumental to responsible policy-making related to the implementation of noninvasive prenatal testing in Canada.

Noninvasive prenatal testing allows genetic testing through the analysis of cell-free fetal DNA circulating in maternal blood. Although not as accurate as diagnostic tests such as amniocentesis or chorionic villus sampling, noninvasive prenatal testing may provide important benefits for pregnant women and their families. In contrast with invasive methods, it carries no risk of miscarriage. In addition, it can be done in early pregnancy (weeks 10–11) and has better detection rates than current first-tier screening methods. The test is thus being gradually implemented worldwide, and is currently available in more than 60 countries,[1] commercially in most, and covered by public funding in some. To date, in Canada, Ontario and British Columbia offer public funding for noninvasive prenatal testing, but only under specific conditions.[2,3] More recently, the Quebec Ministry of Health announced that it will cover the cost of its use in high-risk pregnancies.[4]

Scholars and advocacy groups have expressed concerns regarding the potential consequences of "routinizing" noninvasive prenatal testing.[5,6] These concerns include an erosion of free and informed consent, in addition to the possibility of increased rates of pregnancy termination, which could lead to decreased prevalence of certain genetic conditions and may lead to decreased medical and social support for people and families living with these conditions.[7–10] Thus, it is important to empirically assess the ethical and societal acceptability of noninvasive prenatal testing's clinical implementation and coverage through public funding, to determine that they reflect the interests, needs and values of Canadians.[11] Such decisions should be based on robust evidence regarding cost-effectiveness,[12] and on the views and preferences of stakeholders.[13] In addition, we argue that these decisions should be informed by the evolving perspectives of Canadian experts on ethical and societal issues related to prenatal testing and

Competing interests: None declared.

This article has been peer reviewed.

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CMAJ Open **2018. DOI:10.9778/cmajo.20180083**

genomic technologies. In this study, we aimed to identify the most important benefits, challenges and guiding ethical principles regarding the clinical integration of noninvasive prenatal testing in Canada, according to such experts.

Methods

The Ethical Delphi technique, an "approach for characterizing ethical issues raised by the use of novel biotechnologies," was ideally suited to the aim of this study.[14] The classical Delphi method consists of surveying a panel of experts on a given topic, providing the panel with aggregate results, and obtaining feedback on the results in subsequent rounds. The method promotes an honest, evolving and constructive exchange between people from diverse backgrounds and aims to identify areas of agreement and disagreement regarding emerging areas of knowledge.[15,16] It is a semianonymous method in which research participants are known to investigators but not to one another. Preserving such anonymity provides the respondents with the freedom to change their opinion from one round to the next, rather than defending a locked-in position with their name attached to it.[17] We conducted a 3-round Ethical Delphi study with participants with a diversity of experience with, knowledge of and perspectives on the ethical and societal aspects of prenatal testing and genomic technologies.

Recruitment of the expert panel

We identified people who had been actively involved in previous scholarly (e.g., academic literature) or public (e.g., newspaper articles) discussions on the ethical and societal aspects of prenatal testing and genomic technologies, in addition to relevant professional or advocacy activities in Canada for at least 3 years, as potential participants. We recruited participants from 4 groups: health care professionals, researchers in social sciences and humanities, patient or disability rights advocates, and cultural or religious community advocates. Before completing round 1, participants were asked to confirm that they indeed identified as belonging to one of the 4 groups, and to specify their professional, academic or advocacy expertise. We aimed to recruit 10–15 respondents per group,[17,18] and tried to ensure a response rate of more than 70% in each round to minimize selective participant retention bias.[19,20] Special efforts were made during recruitment to account for language distribution, sex ratio and geographic representation. Participants were invited to participate by email, and were not remunerated for their involvement.

Questionnaires and analysis

In round 1 (May–August 2015), research participants were asked to provide demographic and other relevant information about themselves. Using open-ended questions, we invited them to state, in their own words, the most important "clinical benefits," "ethical, legal, and social issues and concerns" and "moral principles, social norms and values" related to the implementation of noninvasive prenatal testing in Canada. We formulated these questions as broadly as possible to avoid orienting participants' responses from the outset. For the same reason, we deliberately avoided importing concerns from the existing literature at any round of the study. Upon completion of round 1, two reseachers (Birko and Dupras) independently assessed participants' responses and clustered qualitative data using NVivo 10 (QSR International).

Following the process outlined by Burnard,[21] we generated 3 lists of nonredundant benefits, challenges and guiding ethical principles relevant to the implementation of noninvasive prenatal testing in Canada. The second round (January–April 2016) questionnaire was created based on the responses and exact terminology employed by research participants in round 1. Using multiple-choice questions, we asked participants to select the most important benefits, challenges and principles that had been formulated by their peers, according to their own opinion and their estimation of public opinion. The third round (September–December 2016) consisted of an identical set of multiple-choice questions, complemented by a quantitative presentation of the aggregate responses from round 2. Informed by the panel's position, participants had the opportunity to revise or confirm their responses. The 3 rounds were piloted by Haidar and Lemoine for face and content validity.

Ethics approval

We obtained ethics approval from the University of Montréal Comité d'éthique de la recherche en santé (#14-104-CERES-D) before each round.

Results

We identified a total of 302 potential participants, 61 of whom completed round 1 (initial participation rate 20.2%). The makeup of the panel included 17 health care professionals, 17 researchers, 17 patient or disability rights advocates, and 10 cultural or religious community advocates (Tables 1 and 2). Of the 61 experts who completed round 1, 95.1% completed round 2 (n = 58/61), of whom 84.5% completed round 3 (n = 49/58). The resulting overall retention rate was 80.3% (n = 49/61), with rates considerably higher than the 70% per round usually recommended for Delphi studies.[22] Among the participants who completed all 3 rounds, 30.6% were health care professionals (n = 15/49), 28.6% were researchers in social sciences and humanities (n = 14/49), 28.6% were patient or disability rights advocates (n = 14/49), and 12.2% were cultural or religious community advocates (n = 6/49). The initial panel was highly heterogeneous in terms of participants' opinions about the "overall acceptability" of noninvasive prenatal testing in Canada: 55.1% (n = 35/61) perceived the test to be *mostly acceptable* (score 1, 2 or 3 on 7-point Likert scale), and 28.6% (n = 17/61) viewed it as *mostly unacceptable* (score 5, 6 or 7). Such diversity of opinion was important, given our objective of characterizing as many different views and concerns as possible.

Participants identified 10 nonredundant benefits, 27 challenges and 16 guiding ethical principles relevant to the implementation of noninvasive prenatal testing in Canada. Table 3 presents the final lists of prioritized benefits, challenges and principles, as ranked by the panel after round 3.

Interpretation

Throughout the 3 rounds, we paid special attention to reporting the exact terminology employed by participants when stating benefits, challenges and principles. Our study thus has the advantage of offering a detailed landscape of the vocabulary used in practice by experts from diverse epistemologic standpoints. Although many participants assigned value to noninvasive prenatal testing because of its increased accuracy in contrast to current first-tier screening methods, the panel largely agreed that the test's most salient benefits arise when comparing noninvasive prenatal testing to current second-tier diagnostic methods (e.g., no risk of miscarriage, noninvasiveness and results available earlier in the pregnancy). Our study thus confirms the main clinical benefits of noninvasive prenatal testing usually expected and discussed in the literature (safer, easier, earlier).[11]

The panel expected the public to agree with the high ranking of these benefits, but believed Canadians could also be attracted by the idea that the new test provides more information that is potentially useful for decision-making (Appendix 1, available at www.cmajopen.ca/content/6/4/E513/suppl/DC1]). After carefully analyzing challenges and principles, we argue that our study points to 2 sets of requirements that should be prioritized according to Canadian experts: ethical requirements to actively promote free

Table 1: Panel composition

Field of expertise or group represented	No. of participants Round 1	 Round 2	 Round 3
Health care professional	17	16	15
Gynecologist and obstetrician	4	3	15
Genetic counsellor	3	3	3
Neonatalogist	3	3	3
Medical geneticist	2	2	3
Midwife	1	1	1
Nurse	1	2	2
Pediatrician	1	1	1
Other	2	2	1
Social science and humanities researcher	17	17	14
History	4	4	4
Law	3	3	2
Bioethics	2	2	2
Sociology	1	1	1
Anthropology	1	1	0
Philosophy	1	1	0
Other	5	5	5
Patients or disability rights advocate*	17	16	14
Association promoting social values	6	6	6
Association specific to Down syndrome	4	4	4
Association specific to an other condition	2	1	0
Involved in more than one association	2	2	2
Not affiliated to any association	3	3	2
Cultural or religious community advocate	10	9	6
Islam	3	3	2
Christianity	2	2	2
Judaism	2	2	1
First Nations	1	0	0
Other	2	2	1
Total	**61**	**58**	**49**

*We chose not to report the names of affiliated associations to protect the privacy of participants.

and informed decision-making for prospective parents, and societal requirements to protect and promote the rights and interests of vulnerable individuals.

Participants unambiguously stressed how challenging offering adequate counselling to patients can be, and yet largely agreed on the high importance of informed decision-making in the context of prenatal care. Our findings thus resonate with the literature, wherein concerns are expressed as to the potential impact on informed decision-making in terms of implementing noninvasive prenatal testing as "just another blood test"[23–26] and advocating the active promotion of women's reproductive autonomy.[27–30] In addition to protecting free choice, participants underscored the need to ensure the appropriate conditions for evidence-based decision-making. In this respect, they highlighted the difficulty of ensuring the adequate education of health professionals about new prenatal testing technologies and their potential ethical and societal implications. Additional empirical studies will be helpful to identify the needs and preferences of health professionals regarding continuing training.

Table 2: Participant characteristics

Characteristic	Participants, % Round 1 *n* = 61	 Round 2 *n* = 58	 Round 3 *n* = 49
Demographic			
Language*			
English	70.5	70.7	69.4
French	26.2	25.9	26.5
Sex			
Female	55.7	56.9	57.1
Male	44.3	43.1	42.9
Age, yr			
20–39	16.4	17.2	18.4
40–59	49.2	48.3	46.9
60–79	32.8	32.8	34.7
80–99	1.6	1.7	–
Country of birth			
Canada	68.9	67.2	69.4
Province of residence†			
Quebec	39.3	39.6	38.8
Ontario	26.2	25.9	26.5
Alberta	9.8	10.3	8.2
British Columbia	8.2	8.6	8.2
Manitoba	6.6	5.2	6.1
Nova Scotia	3.3	3.4	4.1
Degree completed‡			
Doctoral	60.7	60.3	61.2
Masters	14.8	15.5	18.4
Bachelor	11.5	12.1	10.2
College	11.5	10.3	10.2
Potentially influencing factor			
Having a child living with a disability	23.0	22.4	24.5
Knowing a child living with a disability	86.9	86.2	85.7
Living with a disability	8.2	8.6	8.2
Experienced prenatal screening	47.5	43.1	44.9
Experienced prenatal diagnosis	18.0	17.2	16.3
Disclosed a commercial interest	1.6	1.7	2.0

*Participants speaking other languages accounted for 4.1% at round 3.
†Residents of other provinces or territories all together accounted for 8.2% at round 3.
‡One participant had another level of education but did not complete round 3.

The term "autonomy" did not rank very highly in the final list of principles generated by the participants. This concept may have been perceived as too vague by most participants, or not properly reflecting what is truly at stake with the arrival of noninvasive prenatal testing in Canada. The term "consent" also ranked surprisingly low on the list of challenges, suggesting that although Canadian experts value informed choice, they may believe that procedural modalities of signed consent forms are insufficient to adequately promote it.[31] In the context of noninvasive prenatal testing, promoting reproductive autonomy and protecting consent for prospective parents may instead require enhanced training of health care professionals regarding how best to communicate new testing options to patients, and the importance of understanding individual patients' preferences. Preventing increased pressure to test and pressure to terminate — anticipated consequences of implementing noninvasive prenatal testing in routine prenatal care — is particularly crucial, given our participants' estimation of free choice ranking first according to Canadians (Appendix 1). In the literature, such concerns have consistently been formulated by disability rights advocates, who fear that the emergence of new medical and social norms in prenatal testing may impose coercive pressures on pregnant women to test and terminate affected pregnancies under the guise of "responsible motherhood," thus impeding the voluntary nature of their individual choices.[8,32–36]

Some empirical studies have shown that health care professionals tend to be in favour of testing and terminating affected pregnancies and may thus exercise subtle yet considerable influence on their patient's decision-making process and final choice.[34,37] At the same time, some patients may expect clear recommendations from their health care provider, deliberately and admittedly intending to follow them.[9] To address the imperative of protecting free choice through the provision of relevant, adequate and sufficient information,[38] different strategies exist, such as nondirective counselling (focused on the patient's decisional authority) or shared decision-making (focused on open discussion of personal values and opinions).[39,40] These strategies can increase people's freedom of choice by improving their understanding of available options.[41–43]

More than half of the participants feared that the implementation of noninvasive prenatal testing may lead to increased discrimination against people with disabilities. Many also shared concerns over eugenics and a potential decrease in social support for people with disabilities in the future. They estimated that the threat of eugenics ranks even higher in public opinion (Appendix 1). Perhaps in response to these challenges, the principle of respect for human dignity was perceived by the panel as very important for guiding the implementation of noninvasive prenatal testing in Canada. Many participants underscored the need to continuously promote respect for diversity, and solidarity with individuals living with conditions for which the test screens. These principles point to the societal imperative of being actively committed to the welfare of vulnerable groups. Disability scholarship literature has been useful in highlighting areas of needed improvement in this regard.[44–46]

Although the cost of implementing noninvasive prenatal testing did not rank highly, most participants stressed the importance of ensuring equitable access to the test. Indeed, if it is available only to people who can afford the cost, already vulnerable people or populations could be unfairly disadvantaged by not being offered the same opportunity to manage risks such as miscarriage associated with publicly covered invasive techniques (e.g., amniocentesis) and the perceived burden (e.g., psychological, financial) of raising a child with a severe genetic condition.[40,47] It is worth noting that the cost of implementing noninvasive prenatal testing was estimated to rank first as a challenge in public opinion. However, whether the public is expected to be concerned about the economic burden being imposed on prospective parents (private funding) or on the health care budget (public funding) remains unclear. Empirical studies will be helpful in determining the specific criteria (e.g., level of risk, conditions tested for, funding model) the Canadian public and stakeholders think satisfy the principle of equitable access.

Limitations

We experienced difficulty in recruiting cultural or religious community advocates who had previously expressed their views regarding prenatal testing and genomic technologies. Few such people may have considered themselves sufficiently knowledgeable about noninvasive prenatal testing and thus competent to engage in the study. The resulting lower number of participants

Table 3 (part 1 of 2): Most important benefits, challenges and guiding ethical principles related to the implementation of noninvasive prenatal testing

Factor	Participants, %	Change, % (R3 – R2)
Benefits		
1. No risk of miscarriage	83.7	+13.0
2. Noninvasiveness	63.3	+1.2
3. Results available earlier in the pregnancy	53.1	+6.3
4. Requires only a blood draw	42.9	+5.0
5. Increased accuracy	30.6	+8.2
6. Enhances prospective parents' ability to prepare	24.5	–11.7
7. Decreases anxiety for prospective parents	16.3	–7.8
8. Provides more information potentially useful for decision-making	16.3	–25.1
9. Answers specific needs	10.2	–3.6
10. Potential to expand conditions tested	4.1	–8.0
Challenges		
1. Adequate counselling of patients	79.6	+19.3
2. Pressure to test	67.3	+10.4
3. Adequate education of health professionals	65.3	+10.1
4. Discrimination against people with disabilities	59.2	+10.9
5. Pressure to terminate	55.1	+10.3
6. Eugenics	46.9	+2.1
7. Potential decrease in social support for people with disabilities	44.9	+1.8
8. Routinization of prenatal testing	38.8	+0.9
9. Stigmatization of the parents of children living with the tested conditions	38.8	–0.9
10. Accuracy of the test	34.7	–1.5
11. Decrease in social diversity	34.7	+5.4
12. Adequate professional guidelines	32.7	–5.2
13. Consent	32.7	+1.7
14. Cost of implementation	32.7	–1.8

Table 3 (part 2 of 2): Most important benefits, challenges and guiding ethical principles related to the implementation of noninvasive prenatal testing

Factor	Participants, %	Change, % (R3 – R2)
15. Selective pregnancy termination	32.7	–5.2
16. What conditions to test	30.6	+8.2
17. Increased number of terminated pregnancies	30.6	–0.4
18. Conflicts of interest linked to commercialization	28.6	–2.4
19. Misperceptions of the reliability testing	28.6	–11.1
20. Existence of adequate policies	28.6	–5.9
21. Respect of human rights	26.5	–4.5
22. Access-related issues	24.5	–1.4
23. Increased anxiety for prospective parents	18.4	–7.5
24. Conflict with religious values	14.3	+2.2
25. Assigning responsibility to care for the child	8.2	–0.4
26. Utility of the test	8.2	–12.5
27. Confidentiality of the results	2.0	–13.5
Principles		
1. Informed decision-making	81.6	+2.3
2. Evidence-based decision-making	59.2	+8.9
3. Respect for human dignity	53.1	+2.3
4. Equitable access	51.0	+7.9
5. Free choice	51.0	–0.7
6. Fair resource allocation	46.9	+0.3
7. Respect for diversity	44.9	+1.8
8. Solidarity with people living with the tested conditions	44.9	–10.3
9. Human rights	40.8	+0.6
10. Equality between persons	38.8	+6
11. Nonmaleficence (do not harm)	36.7	–6.4
12. Autonomy	30.6	–7.3
13. Protection of privacy	28.6	–5.9
14. Sanctity of life	28.6	+4.5
15. Inclusiveness	22.4	–6.9
16. Beneficence	18.4	–5.7

in this group, however, should not be perceived as impeding the scientific validity of the results. Our objective was not to compare the views of diverse groups of experts and search for statistically significant associations, but rather to shed light on a diversity of perspectives regarding noninvasive prenatal testing. The specific composition of our panel of experts and the associated limitations should be considered when interpreting the results and their meaning for Canadian policy.

Being pregnant or living with a condition identified through noninvasive prenatal testing were not considered sufficient criteria for participation in our study. Empirical studies with Canadian stakeholders (e.g., PEGASUS surveys involving pregnant women and their partners) will be instrumental in guiding policy-making.[13] However, we did not consider such stakeholders to be experts for the purposes of this study. As reported in Table 2, female participants represented more than half of the panel in each round, with 44.9% of the final panel (*n* = 22/49) having experienced prenatal testing themselves. In addition, considerable numbers of participants reported either knowing a child (*n* = 42/49), having a child (*n* = 12/49) or living themselves (*n* = 4/49) with a disability.

Conclusion

In addition to the ethical and societal considerations discussed here, it is worth noting that some of the issues highlighted by smaller proportions of participants in this study are also informative and should be considered. This is particularly important if we care to consider the voices of people with specific vulnerabilities who, owing to their unique experiences of exclusion, may be more aware of and sensitive to particular ethical and societal issues such as the possibility of decreased social diversity that may follow increased selective pregnancy termination. Potential conflicts of interest linked to the commercialization of noninvasive prenatal testing is another challenge that should be addressed. Otherwise, in the long term, the rise of distrust by some people toward the medical community could further impede effective communication about prenatal testing options. Interdisciplinary appraisals of such issues may be instrumental to responsible policy-making related to the implementation of noninvasive prenatal testing in Canada.

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Contributors: All the listed authors have contributed substantially to this article, approved the final version to be published, and agreed to act as guarantors of the results.

Funding: Fonds du Québec pour la recherche — Santé et Culture provided funding for this project (FQRSC, No. 2014-NP-175854).

Acknowledgements: The authors thank all of the participants involved in this study, as well as Dr. Anne-Marie Laberge and Jessica Le Clerc-Blain for insightful comments and suggestions regarding the design of the study and interpretation of the results. The authors also thank *Fonds du Québec pour la recherche – Santé et Culture* (FQRSC) for providing funding for this project.

Supplemental information: For reviewer comments and the original submission of this manuscript, please see www.cmajopen.ca/content/6/4/E513/suppl/DC1.