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ORIGINAL RESEARCH

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Pregnant women's and policymakers' preferences for the expansion of noninvasive prenatal screening: A discrete choice experiment approach study

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Abstract

Background and Aims: Quantitative approaches for eliciting preferences for new interventions are mostly conducted by patients and rarely by policymakers. This study aimed to quantify the preferences of pregnant women and policymakers regarding the addition of a new test to prenatal screening programs for detecting chromosomal abnormalities.

Methods: A discrete choice experiment was conducted to measure the respondents' preferences for a new prenatal test. A seven-attribute instrument was built based on interviews with pregnant women and policymakers. The data were analyzed using robust conditional logistic regression and nested logit models.

Results: In total, 272 pregnant women and 24 policymakers completed the questionnaire (response rates of 48% and 55%, respectively). Overall, all attributes were statistically significant in the pregnant women group, whereas only three attributes (test performance, degree of test result certainty, and cost) were statistically significant in the policymakers group. Statistically significant differences in test performance and information were observed between the two groups.

Conclusion: Policymakers differed from pregnant women in their appraisal of attributes related to their preference for a new prenatal screening intervention. The low response rates observed in both groups suggest that further investigation of the relevance of this approach must be conducted.

KEYWORDS

discrete choice experiment, health technology assessment, policymakers, pregnant women

What's already known about this topic?

- Noninvasive prenatal screening has the potential to detect various chromosomal anomalies in the fetus, currently non-screened for by public prenatal screening programs.
- Discrete choice experiment (DCE) has been used to better define the characteristics that the primary beneficiaries of a prenatal screening program, the parents, consider relevant to legitimize the addition of a chromosomal condition into the list of conditions sought by the program. Such an approach has never been used for the other main stakeholder, the policymakers who make the final recommendations regarding what to seek in a prenatal screening program.

What does the study add?

- A DCE was undertaken to quantify, by a consensual instrument, the preferences of both pregnant women and policymakers for the addition of a new test into a prenatal screening program to detect chromosomal abnormalities.
- Policymakers did not express a preoccupation with several dimensions of the instrument that were put on the table by the pregnant women, although the corresponding attributes were retained by the policymakers during the instrument construction.

1 | INTRODUCTION

Discrete choice experiments (DCEs) are a preference-based approach that allow quantitative measurement of the relative values that a representative group of the population would assign to different services.[1] The dimensions of a DCE instrument can reflect what the target groups of the population consider important about a health technology. Moreover, once a cost dimension related to technology is included in the instrument, monetary values (i.e., willingness-to-pay [WTP]) can be estimated for the trade-offs that respondents are willing to make to have one unit of change for different aspects of the technology.[2]

Having tools that allow the measurement of patient preferences and demands for health and healthcare interventions is valuable for health technology assessment (HTA) agencies.[3] In each HTA agency (e.g., the National Institute for Health and Care Excellence in England and Wales; the United States Food and Drug Administration; and the Canadian Agency for Drugs and Technologies in Health), these inputs are evaluated by appraisal committees before deliberating national recommendations on the offer of a new intervention. The committee members who are referred to as policymakers consist of diverse experts, such as researchers, clinicians, ethicists, managers, and citizens. This composition aims at capturing the different perspectives required for deliberations.

However, policymakers' concerns regarding technology may differ from those of patients and populations. Indeed, focus group discussions with HTA representatives in Canada, Belgium, and Germany showed that policymakers place more emphasis on information related to the benefits, risks, and administration of the technology.[4] Consequently, clinical efficacy, cost-effectiveness, and equity tend to be the most important factors in funding decisions.[5]

Having an instrument that consensually expresses important dimensions for the beneficiaries of an intervention and policymakers in charge of recommendations regarding the intervention would highlight the fact that decisions made by policymakers on deliberative committees do not necessarily reflect the diversity of opinions and hence, could prove to be a relevant reflection tool for policymakers.

Therefore, we conducted a study using a DCE approach based on a single instrument to measure preferences for an intervention on both the demand and supply sides. The demand group consisted of pregnant women, whereas the supply group consisted of policymakers. The intervention of interest is the expansion of a noninvasive prenatal screening (NIPS) test for the detection of chromosomal abnormalities beyond the currently detected anomalies, which tend to be limited to trisomy 21 (Down syndrome), trisomy 13 (Patau syndrome), and trisomy 18 (Edward syndrome) in most NIPS-based prenatal screening programs.[6–8] The candidates for addition are numerous and have different outcomes and impacts on the life of a child and the family, and the performance of the test for the detection of each condition varies.[7] In a healthcare system that is accountable to the public, the decision to add a new screening condition would ideally consider dimensions that beneficiaries and policymakers consider relevant to the value of the test. In this study, we conducted a DCE study to identify how both groups could converge in determining the characteristics of a new hypothetical chromosomal imbalance detected by the NIPS, which should be considered when deliberating its offer in a public prenatal screening program.

2 | METHODS

Ethical approval for this study was obtained from the two teaching hospitals' ethics committees (*Comité d'éthique de la recherche du CHU de Québec-Université Laval*, project 2020-4877; *Comité d'éthique de la recherche du Centre de recherche du CHU Sainte-Justine*, No. MEO-20-2022-4050, MP-20-2020-4877, Sirul:118984), and permission was granted to enroll pregnant women at the CHUL hospital (*Centre Hospitalier de l'Université Laval*) and the CHUSJ hospital (*Centre Hospitalier Universitaire Sainte-Justine*) in the province of Québec, Canada. Informed consent was obtained from both participant groups (i.e., pregnant women and policymakers) through the online format (by clicking on the "*accept*" function) that gave them access to the questionnaire.

2.1 | DCE development

In this study, a DCE was conducted following the best practice guidelines for conjoint analysis.[9–12] A DCE questionnaire was developed to elicit preferences for a new hypothetical fetal chromosomal imbalance test that could be added to a prenatal screening program currently targeting three common trisomies (T21, T18, and T13). Clinically, numerous fetal chromosomal conditions can be detected by NIPS, including autosomal trisomies, sex aneuploidies, microdeletions, and duplications.[6,8] This technique can even be used to detect mutations.[13] However, the expansion of the screening scope of NIPS beyond the three common trisomies, referred to in this paper as a new hypothetical fetal chromosomal imbalance condition, implies a higher level of uncertainty regarding the phenotype associated with chromosomal abnormality. For example, congenital cardiac anomalies have been reported in approximately 75% of individuals with DiGeorge syndrome (22q11.2 microdeletion) and 50%–75% with 1p36 deletion syndrome.[14,15] Considering the impact of uncertainty in addition to the effectiveness of the test to detect a new condition, the cost of the test, the cost of consequences (not only for the healthcare system but also for families), and the emotional and social impacts of having a child with a chromosomal abnormality for the family, the DCE instrument aimed to measure the preferences of beneficiaries and policymakers for such intervention.

The development of the DCE questionnaire consisted of sequential mixed methods.[16] We conducted a systematic literature review on the use of DCE in prenatal screening for chromosomal anomalies. This review identified potential attributes that have been found to influence preferences for undertaking screening tests for new conditions in a NIPS-based prenatal screening program. The construction of the questionnaire was followed by a qualitative study to test the attributes suggested in the literature and identify others considered important by both groups (i.e., pregnant women and policymakers) regarding the addition of new conditions to a public prenatal screening program for fetal chromosomal anomalies. The methods employed for this qualitative study included individual semi-structured interviews with 12 pregnant women and four policymakers. This initial step resulted in a list of 10 attributes, of which 5 were built based on the responses provided by the participants from both groups. A Delphi study was then conducted with the same participants and ended with a focus group discussion to reach a consensus on attributes and attribute levels. Eight attributes were retained. Finally, a pilot project was undertaken with 33 pregnant women (each completing seven comparison choice tasks) to validate the first consensual version of the DCE instrument and test the feasibility of its administration. Due to the very small pool of policymakers (i.e., individuals who voted as members of HTA deliberating committees) in the province of Québec, Canada, and the desire to administer the final version of the DCE questionnaire, policymakers were not included in the pilot study. This pilot study led to dropping one attribute that failed to attain statistical significance. The final instrument contains seven attributes: "conditions to be screened," "test performance," "moment at gestational age to obtain the test result," "degree of test result certainty to the severity of the disability," "test sufficiency," "the test result presented to pregnant women is about," and "cost related to the test." The details are presented in Table 1.

2.1.1 | Experimental design and construction of choice sets

An unforced choice format was used to design the choice sets. Participants were asked, in each choice set, to choose between three generically labeled options: "test A," "test B," or "Neither" (known as the opt-out option) for their respective choice sets. Tests A and B differed in attribute levels, representing the test detecting a hypothetical chromosomal imbalance that could be added to a public prenatal screening program. Pregnant women were asked which test they would prefer, whereas policymakers were asked which test they would prefer to offer.

The construction of choice sets and subsequent analyses were performed using SAS software (SAS Institute, release 9.4). A fractional factorial design was built instead of a full factorial design.[1] An efficiency design was used to increase the precision of the estimates of the effect of each attribute and maximize the trade-offs between alternatives. Given that there is no prior information available on the magnitude or direction of the parameter estimates, it was assumed to be 0.[10] However, a good efficient design was characterized by the orthogonality (no option dominates another), level balance (all levels of each attribute have an equal frequency), and minimum overlap (there is no overlap in attribute levels).[1,9,17] Besides, to ensure that all attributes are well balanced (level balance) for maximum efficiency, we employed a D-efficiency criterion using the SAS *OPTEX* procedure.[10] The model was thus built based on the consideration of degrees of freedom for the main effects (i.e., direct effect of change in the attribute in the choices) and the two-way interactions.

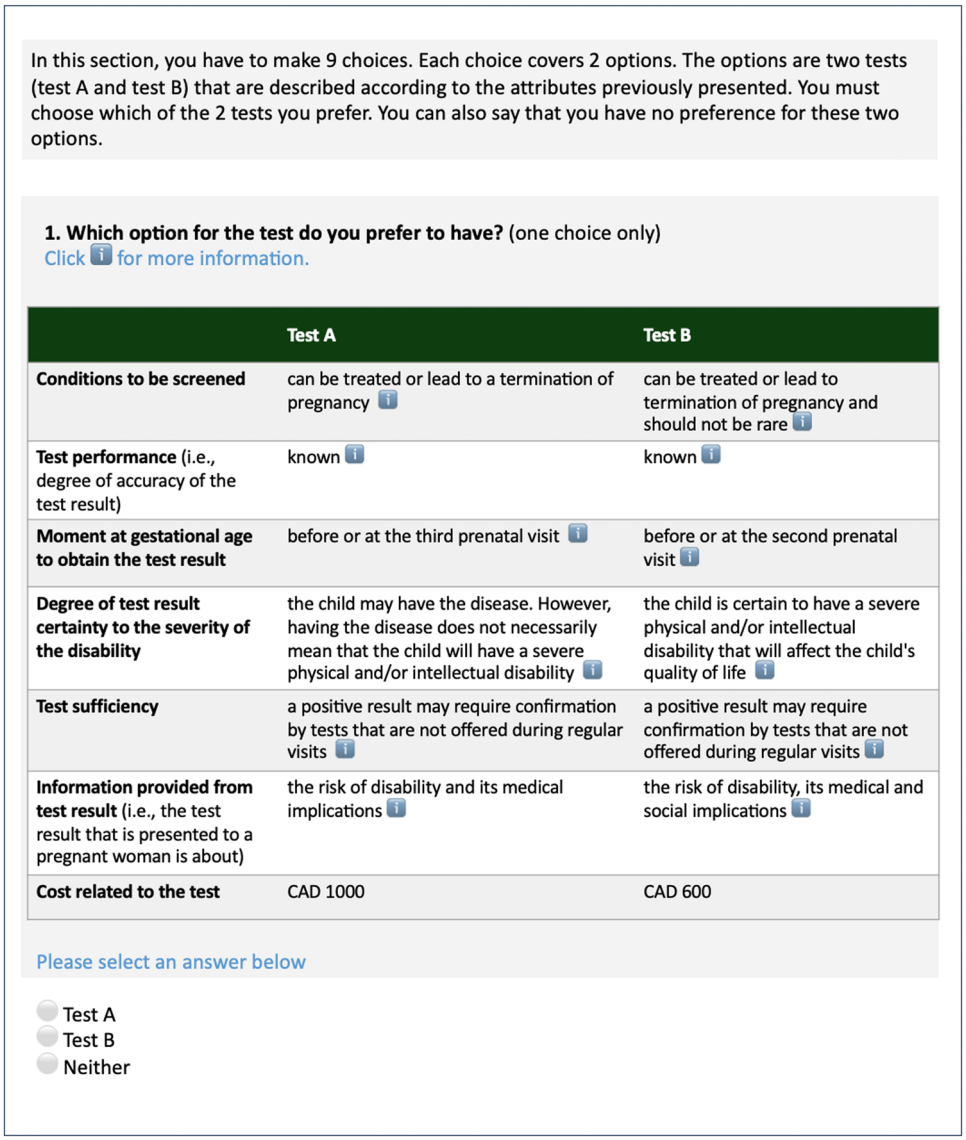
As the ideal number of paired choice sets per respondent remains controversial, it was decided that a different number of choice sets would be given to the two groups of participants (i.e., pregnant women and policymakers) and took into account the sample size constraint.[1,18,19] This also took into consideration the need for sufficient variation in the choice sets and enough choice sets to estimate robust utility values, without exceeding the respondents' cognitive ability.[20] An average of eight choice sets per respondent is generally found in the DCE literature on health problems.[1,17] In the field of antenatal and newborn screening, an average of 11 (range from 4 to 32) has been reported.[21] This study included nine paired-choice sets per pregnant woman and seven sets per policymaker. The choice sets were randomly reorganized to produce several questionnaires. The required sample size was thus defined (detailed in the next section), with one person randomly assigned to each questionnaire.

To test the respondents' understanding and consistency, we used a within-set dominated pair test.[22] In which one of the alternatives was worse than the other with respect to all attributes. Respondents who did not choose the dominant alternative on the test were considered to have failed the consistency test. The results of this fixed question were excluded from the DCE analysis. Response time was used as a proxy for

TABLE 1 List of attributes and levels for DCE survey.

Attributes	Levels	Description
1. Conditions to be screened	1. Can be treated or lead to a termination of pregnancy	A test can detect as many conditions as possible, provided that in case of a positive result, medical intervention is then possible.
	2. Can be treated or lead to termination of pregnancy and should not be rare	A rare disease is defined as a condition that affects less than one in 200,000 individuals. This test would therefore make it possible to detect diseases that are rarer than Down syndrome, which affects 300 children out of 200,000 births.
2. Test performance (i.e., degree of accuracy of the test result)	1. Known	In a few cases, the result of a screening test is incorrect. When the percentage of the error is known, the mother can be told what the probability is, that a second test, called a confirmatory test, which is rarely wrong, will confirm or reject the first test result.
	2. Uncertain	In a few cases, the result of a screening test is incorrect. When the percentage of error is uncertain, the probability that a second, confirmatory test, which is rarely wrong, will confirm or invalidate the first test result cannot be specified. An uncertain result is common for rare diseases.
3. Moment at gestational age to obtain the test result	1. Before or at the third prenatal visit	The result is communicated at the latest during the third prenatal visit, around the 24th week of pregnancy.
	2. Before or at the second prenatal visit	The result is communicated at the latest, during the second prenatal visit, around the 18th week.
4. Degree of test result certainty to the severity of the disability	1. The child is certain to have a severe physical and/or intellectual disability that will affect the child's quality of life	The result may detect a physical or intellectual problem that will lead to a severe disability that will affect the child's quality of life.
	2. The child may have the disease. However, having the disease does not necessarily mean that the child will have a severe physical and/or intellectual disability	The result can detect an intellectual or physical problem but does not indicate the severity of the disability.
5. Test sufficiency	1. A positive result can be confirmed during regular prenatal visits	Screening interventions are offered to all women during a regular pregnancy visit.
	2. A positive result may require confirmation by tests that are not offered during regular visits	Screening interventions may require additional interventions, such as additional visits or specific tests like amniocentesis.
6. Information provided from test result (i.e., the test result that is presented to a pregnant woman is about)	1. The risk of disability	The information is about the possibility that the child may have a disability
	2. The risk of disability and its medical implications	The information is about the possibility that the child may have a disability and the medical consequences of the disability which may require treatment.
	3. The risk of disability, its medical and social implications	The information is about the possibility that the child may have a disability, the medical consequences of the disability, and the social impact of the disability on the life of the child and family.
7. Cost related to the test	1. CAD 0	Out-of-pocket cost for having the test
	2. CAD 200	
	3. CAD 400	
	4. CAD 600	
	5. CAD 800	
	6. CAD 1000	

Abbreviations: CAD Canadian dollars; DCE, discrete choice experiment.



In this section, you have to make 9 choices. Each choice covers 2 options. The options are two tests (test A and test B) that are described according to the attributes previously presented. You must choose which of the 2 tests you prefer. You can also say that you have no preference for these two options.

1. Which option for the test do you prefer to have? (one choice only)
Click for more information.

	Test A	Test B
Conditions to be screened	can be treated or lead to a termination of pregnancy	can be treated or lead to termination of pregnancy and should not be rare
Test performance (i.e., degree of accuracy of the test result)	known	known
Moment at gestational age to obtain the test result	before or at the third prenatal visit	before or at the second prenatal visit
Degree of test result certainty to the severity of the disability	the child may have the disease. However, having the disease does not necessarily mean that the child will have a severe physical and/or intellectual disability	the child is certain to have a severe physical and/or intellectual disability that will affect the child's quality of life
Test sufficiency	a positive result may require confirmation by tests that are not offered during regular visits	a positive result may require confirmation by tests that are not offered during regular visits
Information provided from test result (i.e., the test result that is presented to a pregnant woman is about)	the risk of disability and its medical implications	the risk of disability, its medical and social implications
Cost related to the test	CAD 1000	CAD 600

Please select an answer below

Test A
Test B
Neither

FIGURE 1 A sample of a DCE question. DCE, discrete choice experiment.

cognitive engagement. Data from the pilot project (paper submitted) suggested that the minimum amount of time required to complete the questionnaire was approximately 2 min. Those with a shorter response time and those who took over 24 h were excluded. Figure 1 presents a sample of a DCE question (for more details, see the Supporting information file: Section 4. A blank copy of the questionnaire)

2.2 | Study sample and recruitment

For pregnant women, a fractional factorial design was built with 72 scenarios chosen among the 576 possible combinations, which allows for the estimation of 67 parameters of the model.[23] This criterion ensured that all attributes were well balanced for maximum efficiency and led to a targeted sample size of 284 pregnant women.[10] As for policymakers, since we intended to have a questionnaire version of seven choice sets per policymaker, 210 paired choice sets were randomly organized into 30 blocks of seven. This yielded a target sample of 30 policymakers. Details of the sample size estimation are provided in the Supporting Information.

The participants in this study were pregnant women and policymakers. Pregnant women were eligible for the study if they: (1) were 18 years or older; (2) were enrolled in a clinical trial (PEGASUS-2, ClinicalTrials.gov Identifier: NCT03831256) and had accepted to be solicited to participate in an additional study; (3) had an e-mail address; (4) were in the 26th or 30th week of pregnancy (i.e., after having responded to the last questionnaire of the PEGASUS-2 project); and (5) were able to communicate and read in French. Policymakers were eligible for inclusion if they were either ex- or present members of permanent committees of Health Technology Assessment Agencies in the province of Québec, Canada (i.e., the *Institut national d'Excellence en Santé et Services sociaux*

[INESSS] and the Coordination Committee of the Pre- and Postnatal Programs of the Québec Ministry of Health and Social Services [MSSS]). These committee members were solicited because of their mandate to evaluate all aspects of a new prenatal screening intervention and thus vote for or against how the service should be delivered to the population. The names of policymakers at the INESSS and MSSS were retrieved from committee members listed on official websites or published in official scientific reports made by these organizations related to screening and services provided to mothers and children. Their professional e-mail addresses were retrieved from the internet.

2.3 | Data collection

Choices in the DCE choice tasks were automatically collected by using Université Laval's (Canada) LimeSurvey platform. Eligible participants received an invitation e-mail containing information regarding the objective of the study and a web link that led them to participate. Once the informed consent form was given by clicking on the "*accept*" function, participants had access to the study questionnaire. The information sheet provided the participants with a description of the DCE instrument's attributes and levels. An example of a choice task was also presented, explaining that they would be asked to choose between tests A, B, or neither and that the choice was about a new test for the detection of a hypothetical new fetal chromosomal anomaly. This test is described as a blood test with various degrees of certainty regarding disease detection.

The fetal condition looked for could be rare or not. It can be treated or lead to pregnancy termination. It was also explained that test results could be obtained at different stages of gestation and that a positive result may require further confirmation procedures that are not offered during regular visits. The participants were also informed that the addition of a new test might not be free. The out-of-pocket costs varied between 0 and the maximum amount paid in the private sector in Canada for the detection of chromosomal anomalies not currently listed in a prenatal screening program with NIPS.

2.4 | Statistical analysis

Respondents who failed the within-set dominant pair tests were excluded from the analysis. First, conditional logistic regression models were used to estimate the relative importance (utility) of each attribute for pregnant women and policymakers.[24] The robust sandwich estimates of Lin and Wei[25] were used for the covariance matrix.[25] Standard errors of the parameter estimates were derived using the PHREG procedure in SAS. The models were adjusted using separate and pooled data from pregnant women and policymakers. The models that analyzed separate data allowed us to estimate the relative importance of the attributes for each group in the additional screening test. No interaction terms were included in this model. The Wald test was used to test the significance of the regression coefficients. Another model was built from pooled data with a dummy variable to differentiate between policymakers and pregnant women, as well as the interaction terms between this dummy variable and the attributes. The interaction terms allowed a comparison of preferences between pregnant women and policymakers.

We then assumed that there might be interaction effects due to the opt-out option, as not being screened is a meaningful option in real life. Therefore, we construct a nested logit model using the MDC procedure.[26–28] This model allows the analysis of choices in a two-level decision process. The process assumed that respondents' choices started by choosing between opt-in (either test A or B) and opt-out (neither) options. Test A was then compared with test B if a choice between tests A and B was made.

Marginal WTP values for a one-unit change (i.e., trade-off) in attributes were estimated in separate models based on the coefficient of interest for the negative value of the cost attribute. In a DCE study, individuals are assumed to select the alternative that provides the highest utility (random utility maximization theory).[17] The utility scores are generated using the following formula:

$$V = ß0 + ß1\text{condition} + ß2\text{test\_performance} + ß3\text{result\_moment} + ß4\text{test\_certainty} + ß5\text{test\_sufficiency} + ß6\text{info\_provided\_to\_women} + ß7\text{info\_provided\_to\_women} + ß8\text{cost} + \varepsilon$$

where V is the utility derived from a hypothetical screening test and ε refers to the error

term. All attributes were considered factors (dummy coded) except for the cost related to the test attribute, which was treated as continuous. This establishes the ranking that participants would have in these tests.

All analyses were done at the 0.05 level of significance.

3 | RESULTS

3.1 | Sample characteristics

Invitations were sent via e-mail to 1051 pregnant women and 100 policymakers. Of these, 585 pregnant women and 44 policymakers participated in the survey. The DCE was completed with full responses from 284 pregnant women (response rate: 48%) and 24 policymakers (response rate: 55%). Of these, 12 pregnant women were excluded because they failed the within-set dominated pair test. This group was heterogeneous in sociodemographic characteristics (data not shown).

The demographic characteristics of the participants are presented in Table 2. The mean age of the 272 pregnant women included in the analysis was 31 years (19–39 min), whereas it was 56 years (33–74 min) for the policymaker group. Only four women (1.47%) and three policymakers (12.5%; two women and one man) were single. Of the pregnant women who answered the questions, 87% had a college degree or higher, 90% had a family income of CAD 50,000, 60% were

primigravidas, and 40% knew (either themselves or others) a child born with congenital diseases. All the policymakers who answered the question had a family income of CAD 75,000 or higher.

3.2 | DCE preference results

3.2.1 | Relative importance of attributes on choice

The relative importance (utility) of each attribute for pregnant women and policymakers who expressed their trade-off decisions is presented in Table 3 (model information is provided in the Supporting Information).

Analyses of the pregnant women sample showed that all attributes were statistically significant ($p < 0.05$). Cost related to the test came out as the most important attribute, followed by test performance, degree of test result certainty, information provided from test result, the moment at the gestational age to obtain the test result, test sufficiency, and conditions to be screened as the least important ones. For the most important attributes, pregnant women preferred (compared to the reference attribute level) a test whose performance was known (2.77 times), provided information about the severity of the disability (2.32 times), informed them about the test results on disability risk and its medical and social implications (1.73 and 2.43 times, respectively), and was at a lower cost (0.99 times).

Analyses of the policymakers' sample showed that only three attributes were statistically significant ($p < 0.05$): test performance, degree of certainty regarding the severity of the disability, and cost related to the test. Test performance was considered the most important attribute, followed by the cost related to the test and the degree of test result certainty. Policymakers preferred a test whose performance was known, with certainty about the severity of a disability, and the cost of which was the lowest.

Furthermore, the results for the pregnant women and policymaker samples obtained when applying a nested logit model gave a similar relative importance order of attributes (details provided in the Supporting Information).

3.2.2 | Comparison of preferences between pregnant women and policymakers

The analysis of the pooled samples allowed us to compare the preferences of pregnant women and policymakers. The results are summarized in Table 4. The results show that pregnant women and policymakers differed significantly in the importance given to "test performance" ($p = 0.03$). Policymakers placed more importance on this dimension than on pregnant women (coefficients = 1.643 and 1.019, respectively). Both groups also differed significantly on the information provided from the test result ($p = 0.03$). Women tended to value the completeness of the information provided in the case of a positive screening result, that is, being informed of the expected medical and social outcomes of their child.

TABLE 2 Survey sample demographic characteristics.

	Pregnant women ($n = 272$)	Policymakers ($n = 24$, female: 10, male: 14)
Age, median, years (IQR)	31 (28–33)	58 (45–65)
Marital status, *n* (%)		
Married/live with partner	268 (98.53)	21 (87.5)
Single	4 (1.47)	3 (12.5)
Prefer not to answer	0 (0)	0 (0)
Educational level, *n* (%)		
Secondary[a]	4 (1.47)	-
Secondary School Diploma[b]	7 (2.21)	-
Diploma of Vocational Studies	22 (8.09)	-
College (CÉGEP)	56 (20.59)	-
University	104 (38.24)	-
Master and higher	79 (29.04)	-
Prefer not to answer	1 (0.37)	-
Family incomes (CAD), *n* (%)		
Less than 25,000	2 (0.74)	0 (0)
25,000–50,000	14 (5.15)	0 (0)
50,001–75,000	37 (13.6)	0 (0)
75,001–100,000	56 (20.59)	1 (4.17)
100,001–125,000	51 (18.75)	2 (8.33)
125,001–150,000	57 (20.96)	1 (4.17)
More than 150,000	46 (16.91)	17 (70.83)
Prefer not to answer	9 (3.31)	3 (12.50)
Knowing a child born with congenital diseases, *n* (%)		
Yes	110 (40.44)	-
First pregnancy, *n* (%)		
Yes	179 (65.81)	-
Having children, yes (%)	103 (62.13)	21 (87.50)
1	75 (27.57)	-
2	24 (8.82)	-
3	3 (1.10)	-
4	1 (0.37)	-

Note: Dash "-" in policymakers column indicates that no information was collected.

Abbreviations: CAD, Canadian dollars; CÉGEP, General and professional teaching college (Collège d'enseignement général et professional); IQR, interquartile range.

[a]Classified in Quebec educational system, equivalent to grades 1–6.

[b]Classified in Quebec educational system, equivalent to grades 7–11.

TABLE 3 Robust conditional logistic regression results of discrete choice experiments for pregnant women and policymakers.

		Pregnant women					Policymakers				
Attributes	**Levels**	**Coefficient (95% CI)**	**SE**	**Wald chi-square**	**Pr > ChiSq**	**Odd ratio (95% CI)**	**Coefficient (95% CI)**	**SE**	**Wald chi-square**	**Pr > ChiSq**	**Odd ratio (95% CI)**
Conditions to be screened	1	0.144 (0.036–0.253)	0.056	6.782	0.009	1.155 (1.036–1.288)	0.063 (−0.439 to 0.564)	0.256	0.060	0.81	1.065 (0.645–1.758)
	2	-	-	-	-	-	-	-	-	-	-
Test performance	1	1.019 (0.895–1.144)	0.063	257.832	<0.001	2.772 (2.447–3.139)	1.643 (1.089–2.197)	0.282	33.833	<0.001	5.171 (2.973–8.995)
	2	-	-	-	-	-	-	-	-	-	-
Moment at gestational age to obtain the test result	2	0.584 (0.471–0.697)	0.058	102.466	<0.001	1.793 (1.601–2.007)	0.196 (−0.321 to 0.713)	0.26	0.550	0.46	1.216 (0.725–2.041)
	1	-	-	-	-	-	-	-	-	-	-
Degree of test result certainty to the severity of the disability	1	0.841 (0.723–0.960)	0.061	192.434	<0.001	2.320 (2.060–2.613)	1.243 (0.635–1.850)	0.310	16.057	<0.001	3.464 (1.887–6.362)
	2	-	-	-	-	-	-	-	-	-	-
Test sufficiency	1	0.433 (0.323–0.542)	0.056	60.000	<0.001	1.541 (1.382–1.720)	0.458 (−0.004 to 0.919)	0.235	3.783	0.052	1.580 (0.997–2.506)
	2	-	-	-	-	-	-	-	-	-	-
Information provided from test result	2	0.548 (0.410– 0.687)	0.071	60.294	<0.001	1.731 (1.507–1.988)	−0.241 (−0.938 to 0.457)	0.356	0.457	0.50	0.786 (0.391–1.580)
	3	0.892 (0.750–1.033)	0.072	152.893	<0.001	2.439 (2.118–2.809)	0.438 (−0.124 to 1.000)	0.287	2.337	0.13	1.550 (0.884–2.719)
	1	-	-	-	-	-	-	-	-	-	-
Cost related to the test	one unit ascension	−0.002 (−0.002 to −0.001)	0.0001	277.932	<0.001	0.998 (0.998–0.999)	−0.002 (−0.003 to −0.001)	0.0004	31.569	<0.001	0.998 (0.997–0.999)

Abbreviations: CI, confidence interval; SE, standard error.

TABLE 4 Preference differences between pregnant women and policymakers on the screening test (robust conditional logistic regression).

		Pregnant women		Policymakers		Comparison		
Attributes	**Levels**	**Coefficient**	**SE**	**Coefficient**	**SE**	**Deviation**	**SE**	**Pr > ChiSq**
Conditions to be screened	1	0.144	0.055	0.063	0.256	0.082	0.262	0.76
	2	-	-	-	-		-	-
Test performance	1	1.019	0.063	1.643	0.282	**−0.624**	0.290	**0.03**
	2	-	-	-	-		-	-
Moment at gestational age to obtain the test result	2	0.584	0.058	0.196	0.264	0.388	0.270	0.15
	1	-	-	-	-		-	-
Degree of test result certainty to the severity of the disability	1	0.841	0.061	1.243	0.310	−0.401	0.316	0.20
	2	-	-	-	-		-	-
Test sufficiency	1	0.433	0.056	0.458	0.235	−0.025	0.242	0.92
	2	-	-	-	-		-	-
Information provided from test result	2	0.548	0.071	−0.241	0.356	**0.789**	0.363	**0.03**
	3	0.892	0.072	0.438	0.287	0.453	0.296	0.13
	1	-	-	-	-		-	-
Cost related to the test	One unit ascension	−0.0015	0.0001	−0.0021	0.0004	0.0006	0.0004	0.13

Abbreviation: SE, standard error.

TABLE 5 Marginal willingness-to-pay for changes in attributes (CADs).

		Opt-out option excluded		Opt-out option included	
Attributes	**Levels**	**Pregnant women (95% CI)**	**Policymakers (95% CI)**	**Pregnant women (95% CI)**	**Policymakers (95% CI)**
Conditions to be screened	1	95.67* (22.19–169.14)	29.90 (0–269.79)	159.83* (60.80–258.86)	48.36 (0–330.00)
Test performance	1	675.14* (546.51–803.77)	782.80* (341.07–1224.52)	778.20* (612.17–944.23)	788.60* (221.37–1355.83)
Moment at gestational age to obtain the test result	2	386.52* (294.75–478.31)	93.32 (0–340.94)	440.90* (316.01–565.79)	142.67 (0–436.02)
Degree of test result certainty to the severity of the disability	1	557.26* (447.31–667.20)	591.98* (179.13–1004.83)	594.28* (451.15–737.42)	585.53* (114.65–1056.42)
Test sufficiency	1	286.56* (202.94–370.17)	217.98† (0–451.21)	311.16* (200.39–421.94)	272.659‡ (0–601.04)
Information provided from test result	2	363.21* (259.27–467.16)	-114.6486 (0–220.36)	410.78* (268.63–552.94)	91.01 (0–435.74)
	3	590.46* (464.39–716.53)	208.82* (0–501.44)	645.84* (473.24–818.43)	392.47* (0–785.95)

Note: Opt-out option excluded: Robust conditional logistic regression model; Opt-out option included: Nested logit model.

*Denotes significance at the 5% level.

†p-value = 0.0518.

‡p-value = 0.0556.

3.3 | Marginal WTP and utility score

Table 5 shows that differences between pregnant women and policymakers can be observed through WTP values. Policymakers and pregnant women might be willing to pay different amounts for knowing more about the test performance and the degree of test result certainty relative to the severity of the disability. However, the confidence intervals overlapped, making it difficult to draw definitive conclusions.

TABLE 6 Utility scores of different tests by pregnant women and policymakers.

	Attribute levels							Utility score	
Test	Conditions to be screened	Test performance	Moment at gestational age to obtain the test result	Degree of test result certainty	Test sufficiency	Information provided from test result	Cost related to the test	Pregnant women	Policymakers
1	1	1	2	1	1	2[a]	1	3.57	-
2	1	1	2	1	1	2	2	3.27	-
3	1	1	2	1	1	2	3	2.97	-
4	1	1	2	1	1	2	4	2.66	-
5	1	1	2	1	1	2	5	2.36	-
6	1	1	2	1	1	2	6	2.06	-
7	1	1	2	1	1	3[b]	1	3.91	-
8	1	1	2	1	1	3	2	3.61	-
9	1	1	2	1	1	3	3	3.31	-
10	1	1	2	1	1	3	4	3.01	-
11	1	1	2	1	1	3	5	2.71	-
12	1	1	2	1	1	3	6	2.40	-
13	-	1	-	1	-	-	1	-	2.89
14	-	1	-	1	-	-	2	-	2.36
15	-	1	-	1	-	-	3	-	1.84
16	-	1	-	1	-	-	4	-	1.31
17	-	1	-	1	-	-	5	-	0.79

[a]Holding "Information provided from test result" attribute at level 2, Utility W2 = 0.14 + 1.02 + 0.58 + 0.84 + 0.43 + 0.55 + 0 − (0.0015 × cost)
[b]Holding "Information provided from test result" attribute at level 3, Utility W3 = 0.14 + 1.02 + 0.58 + 0.84 + 0.43 + 0 + 0.89 − (0.0015 × cost)
Utility PM = 1.64 + 1.24 − (0.002 × cost).

Using the coefficients presented in Table 3, the utility scores of the hypothetical examples of screening tests with different attributes were generated (Table 6). As all attributes were significant to the pregnant women's choice, the scores were generated by holding on to the information provided from test result by changing between levels 2 and 3. For policymakers, scores were generated based on only the three attributes that were significant to their choices.

The results suggest that when test result information about the risk of disability and its medical complications is withheld, pregnant women will place the greatest utility (3.57 for Test 1) on a test that is free of charge. When information about social complications of disability was added, they obtained the highest utility score (3.91 for Test 7). For policymakers, when the cost attribute levels changed, the free test had the greatest utility (2.89 for Test 13).

4 | DISCUSSION

This study aimed at measuring the preferences of pregnant women and policymakers for the addition, to a prenatal screening program, of a new test allowing the detection of chromosomal abnormalities other than the three common conditions (trisomies 21, 13, and 18). This was done using a single DCE instrument specifically developed to measure the preferences of pregnant women and policymakers. The instrument allows the identification of differences between both groups in terms of the importance of attributes consensually defined by both groups, which should be considered when a decision is made regarding the eventual addition of new conditions to screen for.

To our knowledge, this is the first study to use a common DCE instrument to quantify values put forth by both sides of the healthcare system (i.e., beneficiaries and policymakers) who directly influence the offer of services (i.e., prenatal care intervention) to the population. Moreover, previous DCE studies on prenatal screening programs have focused on pregnant women, partners, or health professionals and have not yet considered policymakers.[29–33]

In the pregnant women group, all attributes were statistically significant, denoting their importance. This result was not surprising because pregnant women participated in the construction of the DCE instrument (paper under review). Among these attributes, they placed the greatest value on the out-of-pocket money to pay for the test, followed by test performance. These findings might be

slightly different from those of other DCE studies on antennal care, in which the accuracy of the test tended to be commonly reported as the most important attribute. In the present study, this observation may reflect the fact that the study took place in Canada, where healthcare services defined as medically necessary are paid by the government without patients' out-of-pocket contributions. Therefore, the population is unfamiliar with paying for it. In this regard, we note that economic evaluations conducted in Canada have been suggested to reflect the funding basis of the system.[34] WTP studies performed in the country are therefore expected to avoid considering out-of-pocket expenses for non-medically necessary services.[35,36] However, one of the objectives of the study was to be able to calculate WTP. Therefore, we included an out-of-pocket attribute.

In the policymakers group, not all attributes were statistically significant. Only three attributes were identified: the test performance, degree of certainty provided by the test results regarding the severity of the disability, and the cost related to the test. The "test sufficiency" attribute whose p-value ($p = 0.052$) was close to the significant threshold. We cannot discard the possibility that if the targeted sample size had been reached, this dimension would have come out statistically significant too.

The first three attributes (performance, certainty, and sufficiency) were as expected, as they reflect the main issues related to the effectiveness of an intervention that is usually discussed in deliberating committees. The last attribute (cost) was also as expected, as policymakers in Canada tend to be concerned with the expected financial burden for patients and the population brought about by an innovation that would be introduced into the healthcare system. Their decisions in deliberative committees must be consistent with the fundamental principle of the Canadian healthcare system, which is a system where the consumption of necessary medical services is completely free of charge.[37]

However, other attributes of the DCE instrument were not significant for policymakers. This might be the result of a consensus between pregnant women and policymakers over the dimensions that should be included, which was inferred throughout the instrument's construction. It is possible that policymakers felt that an attribute presented as important by women but which they did not consider essential should not be rejected. In addition, we cannot rule out the possibility that the non-significance of certain attributes is because of the poor statistical power.

Finally, our findings support evidence from the literature that policymakers tend to make decisions considering several aspects that may be of little importance to other groups, including beneficiaries.[38–41] Yet, most studies on this question were performed using qualitative approaches. There is some added value to examining the differences in the values given by different stakeholders with a common quantitative instrument. A common DCE instrument can provide quantitative data that expresses the relative importance of attributes. As such, the instrument forces the reflexivity of the deliberative committees' capacity to reflect the concerns of patients and the population.

4.1 | Study limitations

This study has some limitations.

First, the response rates of pregnant women and policymakers did not differ significantly. In particular, approximately half of the policymakers who agreed to participate did not complete the questionnaire. Participants from both groups stopped answering after the first question. Although the reasons for the refusal of potential participants were not investigated, some policymakers provided their feedback (via answering invitation e-mail) on the questionnaires. The main reasons mentioned were being uncomfortable with some of the trade-off questions, having to deal with a complicated study subject, having to answer a questionnaire that felt too abstract, and finding the questionnaire unable to address how they perceived the problem. Further investigations exploring policymakers' acceptance of the DCE approach are thus warranted.[42]

Second, we must acknowledge that being unable to reach the policymaker group's targeted sample size might have affected our statistical capacity to observe changes between pregnant women and policymakers and their WTP values. The response rates may also have influenced the values associated with each attribute provided by both groups.[43] This could also limit the generalization of the study results to different contexts.

Third, the D-optimal criteria were minimized in the experiment design for the policymakers group due to the sample size constraint. This may have limited the capacity of the DCE to predict policymakers' choices.[18] However, the fact that results obtained from policymakers can be explained might effectively reflect their preferences.

Finally, only a within-set paired test was used in the present study to check internal validity. Other tests have been proposed by DCE builders[22,44] including tests for attribute dominance, which indicate the extent to which respondents focus on one attribute and almost always select the alternative with the best level of that attribute. In our study, further analysis of the respondents who failed the validity test suggested that they did not pay attention to the content of the questions.

5 | CONCLUSION

This study shows that the use of a consensual DCE instrument built by the beneficiaries of a health intervention and policymakers provides different responses when applied to representative samples from both groups. Policymakers did not express a preoccupation with several dimensions of the instrument that were put on the table by the patients, despite the fact that the corresponding attributes were retained by policymakers during instrument construction. This raises questions about the capacity of an instrument such as the DCE to increase the involvement of the patient/population perspective in a decision-making process related to the provision of health services in a public health system.

AUTHOR CONTRIBUTIONS

Hung Manh Nguyen: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; writing—original draft; writing—review and editing. **Mohammad Baradaran**: Data curation; project administration; resources; software; visualization. **Gaétan Daigle**: Data curation; formal analysis; methodology. **Leon Nshimyumukiza**: Writing—review and editing. **Jason Robert Guertin**: Writing—review and editing. **Daniel Reinharz**: Supervision; Writing—review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data supporting the findings of this study are available from the corresponding author upon reasonable request and through supplementary documents.

ETHICS STATEMENT

Ethics approval for this study was obtained from two teaching hospitals' ethics committees: *Comité d'éthique de la recherche du CHU de Québec-Université Laval* (project 2020-4877); *Comité d'éthique de la recherche du Centre de recherche du CHU Sainte-Justine* (No. MEO-20-2022-4050, MP-20-2020-4877, Sirul: 118984). Permission was granted to enroll pregnant women at the CHUL hospital (*Centre Hospitalier de l'Université Laval*) in Québec City (Canada) and at the CHUSJ hospital (*Centre Hospitalier Universitaire Sainte-Justine*) in Montréal City (Canada). Informed consent was obtained from the participants through the online format that gave access to the questionnaire.

TRANSPARENCY STATEMENT

The lead author Daniel Reinharz affirms that this manuscript is an honest, accurate, and transparent account of the study being reported; that no important aspects of the study have been omitted; and that any discrepancies from the study as planned (and, if relevant, registered) have been explained.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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