Development of a Discrete Choice Experiment (DCE) questionnaire to elicit values by pregnant women and decision-makers for the expansion of a NIPS-based prenatal screening program

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Research

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Abstract

Background

In an accountable world, being able to take into account the value given by relevant stakeholders to an intervention that could be offered to the population is considered as desirable. DCE is an approach particularly suited for the measurement of such values in the field of prenatal care. Yet, DCE studies in the field of prenatal screening have focused mainly on pregnant women and their care providers but have neglected another key actor, the decision-makers. The objective of the study was to develop a DCE instrument applicable to pregnant women and decision-makers, for the evaluation of new conditions to be added to a screening program for fetal chromosomal anomalies.

Methods

An instrument development study was undertaken. Methods employed included a literature review, a qualitative study performed on pregnant women and decision-makers, and a pilot project to validate the developed instrument and test the feasibility of its administration through an online survey platform.

Results

An initial list of ten attributes and levels were built from the information provided by the literature review and the qualitative research component of the study. Seven attributes were built based on responses provided by participants from both groups. Two attributes were built from what was said by women only and one from what was said by decision-makers only. Search for consensus through consultations and a focus group discussion led to the retention of eight attributes. A pilot project was then performed with 33 pregnant women. This led to the exclusion of one attribute that showed poor influence on the choice making. The final version of the instrument contains seven attributes.

Conclusion

This paper presents the construction of a DCE instrument that can be administered to pregnant women on the demand side, and decision-makers on the supply side. Such an instrument to measure the social desirability of an intervention could be an added value to the decision-making process of Health Technology Assessment agencies.

1. Introduction

Allowing stakeholders such as the population to influence, directly or indirectly, policy decisions is a fundamental component of democratic jurisdictions (1). In the health care sector, this influence is generally referred to by the terms 'participation', 'involvement' or sometimes 'engagement' (2). They cover the multiple modalities that allow different stakeholder groups to bring their input into the decision-making process on what services to provide to the population (3).

Health economic evaluations (HEE) are a methodological approach that allows putting a judgement on the economic value of a health intervention (4). A lot of efforts have been deployed to allow some participation of the public in the HEE process (5–7). This has led to two approaches that include inputs from either patients or the general population, in the estimation of a benefit of having access to a health intervention: cost-benefit analyses (CBA) (via the use of willingness-to-pay and willingness-to-accept analyses) and cost-utility analyses (CUA) (8). Willingness-to-pay (WTP) analyses estimate health benefits as a monetary value whose importance reflects the value given by the respondents to a survey, for the evaluated intervention. Validity and ethical issues make this approach difficult to apply (9). It is therefore seldom used. The second approach, named cost-utility analyses (CUA), uses an indicator, quality-adjusted life-years (QALYs), to express the benefit of an intervention. QALYs quantify health outcome provided by an intervention in terms of gain in length of life adjusted by its patients or general population-defined utility score or desirability (10, 11).

A key issue faced by these approaches is related to the fact that different groups of the population might value an intervention or the health produced by the intervention differently (12)(13). Although CBA and CUA could take into consideration inputs of different groups of the population, they commonly focus on only one group, usually the beneficiaries of the intervention under evaluation (13, 14). Researchers who use these approaches seldom consider other groups, like experts who are members of Health Technology Assessment (HTA) agencies committees and who make final recommendations to policymakers about the adoption of a technology. These experts can value the technology differently compared to the population, based on considerations, which are also socially relevant. This observation mainly comes from the fact that the WTP and utility instruments are built to express the value given to an intervention by its potential or effective beneficiaries (8).

The issue of participation has become even more complicated in the field of prenatal screening for fetal conditions by the fact that utilities cannot be estimated by the fetus (15). Mother's utilities can be used instead, but this would contravene with the habit to consider utilities of the main beneficiary of an intervention.

Thus, seeking for an alternative modality facilitating this participation becomes more desirable in the field of evaluation of prenatal screening programs, as major advances in prenatal screening and diagnosis technology leads to difficult decisions regarding which services to offer to the population. A relatively new blood test, namely non-invasive prenatal screening (NIPS) test, allows to measure analyzing cell-free fetal DNA (cfDNA) in maternal blood. It allows detecting a huge array of chromosomal anomalies (16), besides anomalies currently detected, which tend to be limited to trisomy 21 (Down syndrome), trisomy 13 (Patau syndrome) and trisomy 18 (Edward's syndrome). Decisions regarding which conditions could be added to a public program have to take into consideration, not only the uncertainty of test performance in detecting these new conditions, but also some key ethical, social and economic consequences for the society (17, 18). Very few studies have been done to estimate the expected cost-benefit of expanding the scope of NIPS beyond its use to detect the most common anomalies (i.e., trisomy 21, 18 and 13). For these reasons, there is, in the world of economic evaluation of

prenatal testing, an interest to have methodological approaches that could overcome the limitation of CUA.

A discrete choice experiment (DCE) is a method used to explore the relative importance of different attributes within a decision-making process (19). DCE allows measuring a perinatal intervention that concerns a fetus or a newborn from the adult perspective, solving the problem of the impossibility to define utility values for a fetus (20, 21). A DCE instrument could also reflect the desirability of a perinatal intervention from the perspective of the population/patients and of other groups, as decision-makers or those who work in HTA agencies. It is on the last aspect particularly that this work is focused on.

This paper reports the development of a DCE instrument to measure the preferences of pregnant women and decision-makers for the expansion of the NIPS test for the detection of fetal chromosomal anomalies in a public prenatal screening program.

2. Methodology

The development of the DCE questionnaire was undertaken in sequential steps (Fig.1) based on best practice guidance (19,22–24). The methods and approach for analysis for each stage are summarized in the following sections.

Stage 1: Attribute Development

Attribute development consisted of two distinct steps: (1) attribute identification; (2) attribute selection and framing.

- **Attribute identification**

The process of attribute identification started with a review of literature followed by in-depth interviews with pregnant women and decision-makers.

A systematic review of literature conducted by two researchers (HMN, BS) on the use of DCE in the field of prenatal screening for chromosomal anomalies was performed. The search strategy was validated by a librarian. Data extracted on the identification of attributes and levels for preferences of prenatal screening are presented in additional file 1 (full-unpublished report is available on request). The review led to identifying potential attributes that have been found to influence preferences to undertake a screening test for a new condition in a NIPS-based prenatal screening program.

A qualitative study was then undertaken to test the attributes suggested from the literature and identify others that would be regarded by both groups, the group of pregnant women and the group of decision-makers, as important to consider in the decision-making process regarding the content of a public prenatal screening program.

Semi-structured interviews, based on an interview guide (see additional file 2), were conducted with pregnant women ($n=12$) and decision-makers ($n=4$). This guide had been beforehand pretested with two pregnant women and one decision maker. These three persons were not included into the sample for this project. They were only asked to comment on the understanding of the interview guide.

Inclusion criteria for pregnant women were to be primigravida, aged 18 years or above, and being consulting at the obstetric department of the CHUL hospital in Quebec City (Canada) for a first prenatal echography. Inclusion criteria for decision-makers were to be either member of a permanent scientific committees of the Quebec's HTA Agency, INESSS (*Institut National d'Excellence en Santé et Services Sociaux*), or a public servant at the Ministry of Health and Social Service (Québec), and to be involved in decision-making processes on services to be provided to mothers and children.

All interviews were digitally recorded. The verbatim were transcribed by using NVivo Transcription (QRS International 2020). The transcriptions were independently checked by two researchers (HMN, CL) while relistening to the recordings.

The analysis was independently performed by these two researchers (HMN, CL). It aimed at identifying key attributes and their levels. The initial framework used to guide the identification of the attributes was based on the interview guide (deductive approach) (25,26). Additional codes were generated where required (inductive approach) (25,26).

The analysis started with five pre-established dimensions that were expected to come out from the interviews: 1) monthly family expenses associated with a child's disability; 2) prevalence of a new disease added to the list of diseases searched for by NIPS; 3) performance of the test for this new disease, i.e., probability of identifying a child with a disability; 4) probability that a child tested positive has a severe phenotype; 5) out-of-pocket cost associated with being tested. New dimensions suggested by the interviews were added. Interviews were built in such a way that, at the beginning of the interview, respondents were incited to say whatever they thought was an important characteristic to consider when deciding which condition to screen for, without being interrupted or influenced.

The analysis was first based on triangulation (i.e., categories and themes have to be derived from several sources of information) (27). Moreover, all interviews were coded independently by the two researchers. In case of disagreement, discussions were held until a consensus on the final coding was reached.

Attributes identified by the two groups of respondents were merged. All possible levels arising from the interview were retained. Attributes defined by only one group were also included in the list of attributes/levels for the following selection procedure.

- **Attribute selection and framing**

Attribute selection followed an iterative process aiming at finding a consensus between both groups of participants, on a set of attributes to be included in the DCE questionnaire. The iterative approach was

based on consultations and a focus group discussion. Discussions were held between research team members after each step to refine the list of attributes/levels.

Firstly, the consultation process was undertaken with the same participants who had participated in the attribute identification step and had accepted to be contacted for future study. A list of potential attributes and levels retrieved from the previous codification procedure was presented to the participants. This consultation process aimed to refine the first list of potential attributes and levels. This list was sent to each participant via e-mail, to explore their opinion regarding the dimensions' meaning and relevance. Participants were also asked to provide justification in case they considered one of the attributes to be irrelevant. Attributes that were considered relevant by the participants were thus retained and modified if needed, while those considered irrelevant were excluded.

At the second step, the list of retained attributes was refined based on a focus group discussion conducted with three pregnant women and one decision-maker solicited from the same hospital (focus group discussion with decision-makers alone were not held due to the limited pool of potential participants). They were asked to give their opinion on the relevance of attributes and levels.

Information gained from the attribute-selection process was synthesized by one researcher (MHN). The content of attributes and levels was then revised by the research team members to ensure their relevance and the comprehension of the wording.

Eight key attributes and their levels were identified.

Stage 2: Experimental design and construction of tasks

DCE design and construction of tasks followed the 10-points checklist best practice guidance for conjoint experiment design proposed by the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) (22).

- **Construction of choice tasks**

An experimental design was performed in order to generate unforced choice questionnaires (28). The questionnaires consist of a series of two options that differ by their level at each of the questionnaire's dimensions (i.e., attributes). The attributes represent characteristics of a hypothetical additional test that could be added to the list of tests already included in a public prenatal screening program. Respondents must choose the preferred option or declare that they cannot decide which of the two options is the preferred one.

- **Experiment design**

The number of possible combinations (so-called alternatives) is determined by the number of attributes and levels. This experiment setup involved eight attributes (five attributes with two levels; three attributes with three levels), for a total of $2^5 \times 3^3 = 864$ screening combinations in the full factorial design (29).

Since this number was too high, we built a fractional factorial design using efficient design (instead of orthogonal design due to its limitations of interaction effect estimations), to reduce the sample of scenarios to a manageable level, while still being able to evaluate the effect of each attribute and their 2-way interactions (28,30). The design allowed defining the scenario based on 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) (22,28,30).

1. Design of the questionnaire

Generic labels were used to identify the options, called test A and test B across all the choice sets, because they do not reflect exact screening options and there was a possibility that labels might encourage the use of a heuristic approach (31). An opt-out option (no preference for option A or B) was added although there is risk that an opt-out option could lead to high levels of non-response where a trade-off is judged to be difficult (32). Test A and B were constructed in such a way that trade-offs were expected.

Questionnaires are built in such a way that pregnant women are asked which prenatal test they would prefer to have, whereas decision-makers are asked which prenatal test they would prefer to offer to pregnant women.

Stage 3: DCE survey design

Survey components include an introduction to the survey and explanation on its purpose, consent form, an explanation of attributes and levels, an example of DCE choice task followed by the eight/seven DCE choice tasks (for pregnant women and decision-makers, respectively), and questions on demographic data (age, sex (for decision-makers only), income) and on experience of having or knowing a child with disability.

The DCE questionnaires were transferred on Université Laval's (Canada) LimeSurvey platform (version 3.23.6+200929; <https://www.questionnaires.cstip.ulaval.ca/>).

Stage 4: Pilot testing the DCE Survey

A pilot project was undertaken to explore the feasibility of the survey that will be administered to a large sample of respondents (i.e., explore the understanding of the tasks, the complexity of the choices, the time needed to fill the questionnaire), as well as the statistical relevance of the dimensions and levels of the first version of the final questionnaire. For practical reasons (a limited pool of potential respondents in the group of decision-makers), we could only test the questionnaires through this pilot project with pregnant women.

The pilot project was also used to elicit a dominant choice that will be added in the full-scale study with pregnant women and decision-makers.

- **Sampling and recruitment**

A D-optimal design (33–35) was constructed by judiciously selecting 22 screening combinations, and thus conducted to a pairwise-DCE pilot project (2 by 2 scenarios per choice task) of $C(22,2)=231$ pairs. This project was undertaken using 33 individuals who were asked to give their opinion on 7 different choice tasks ($33 \times 7 = 231$). Detail on sample's calculation of this pilot project can be found in the additional file 3.

Participants had to be at a gestation age between 28 and 30 weeks. They were recruited among participants to a clinical trial (PEGASUS-2, ClinicalTrials.gov Identifier: [NCT03831256](#)) who had accepted to be solicited to participate in an additional study. In this stage, participants were asked to choose the screening option that either suits them the best in each of the tasks or state that they could not express a preference.

- **Data collection and analysis**

Choices in the DCE choice task were collected automatically by the LimeSurvey platform. Eligible participants received an invitation message containing information regarding the nature of the study, and a link that led them to participate in the study. Once the informed consent form was given by clicking on the “*accept*” function, participants had access to the DCE survey.

Participants were given two weeks to answer the survey. After two weeks, the link to the non-answered questionnaire was inactivated and sent to new solicited participants until the sample size had been reached.

The preference data were codified on Excel for analysis using a conditional logit model (SAS the LOGISTIC procedure, release 9.4) to estimate the effects of each attribute on the preference of the participants. The significance level was fixed at 0.25% (36).

3. Results

Stage 1: Attribute Development

Attribute identification methods resulted in the identification of ten categories that could potentially become attributes of a test that should influence, according to the respondents, the decision to expand the list of conditions screened for in a NIPS-based prenatal screening program. Each category contained multiple decisional points suggesting potential levels for each attribute. The source of each category is provided in Table 1. This step showed that pregnant women and decision-makers have many common preoccupations besides specific ones.

Table 1
Decisional points to inform potential attributes with an indication of the sources

Category	Decisional points	Sources
(1) Conditions being screened	Conditions that women are interested to know	DM
	Detect as many as possible conditions	PW
	Detecting severe conditions/anomalies	DM, PW
	The diseases can be confirmed	DM
	Conditions that could also be screened at newborns	DM
(2) Information from the test	Reliable and comprehensive information	DM, PW
	Easy to interpret the results	DM
	Simple information	PW
(3) Test performance	Low false positive (less than 5%) and false negative rates	DM, PW
	Accept higher false positive rate	DM
	Take the test regardless detection rate to reduce risk of any possible problems to the baby	PW
(4) Test procedure	Simple and rapid	DM
	Women' stress	DM
(5) Cost associated with the test	Reasonable cost accepted by women	DM
	At a reasonable cost	PW
	Publicly funded tests	DM
(6) Test uncertainty implication	Women stress/anxiety and public system cost	DM
	Difficult to make confirmation and prognosis	DM
(7) Acceptance of uncertainty level related to physical and intellectual problems	Not accept low level of certainty	PW, DM
	Depend on individuals	DM
(8) Geographical assess to the test	Test can be accessible in the proximity, either hospitals or clinics	PW
(9) Moment in gestational age to obtain test result	As soon as possible	PW
	8 - 10 weeks	PW

(PW-pregnant women; DM-decision-makers)

Category	Decisional points	Sources
	12 weeks	PW
<i>(PW-pregnant women; DM-decision-makers)</i>		

Indeed, several preoccupations were shared by both groups, notably regarding the likely need to select which conditions in the list of chromosomal anomalies that can be screened for by NIPS, the type of information resulting from the test that should be transmitted to parents, the test performance, the parent's out-of-pocket costs related to the test, and the level of certainty of the test result. Preoccupations regarding these categories could differ between pregnant women and decision-makers. For example, pregnant women desired to avoid excluding conditions, while decision-makers were concerned with limiting the choice to conditions whose disability can be prevented or mitigated thanks to a medical intervention. Sensibility and specificity (false negative rate and false positive rate) issues were raised by all four decision-makers, but not by pregnant women. Decision-makers, unlike women, did not raise the question of gestational age to receive the result of the test, and the question of 'geographical access to the test'. They tended to place greater emphasis on the complexity of the test procedure and discussed the issue of stress that the test might cause to women. Details are provided in Table 2.

Table 2
List of potential attribute and levels

Pregnant Women		Decision-Makers		Consensus	
Attributes	Levels	Attributes	Levels	Attributes	Levels
Conditions to be screened	1. Detect as many as possible conditions	Conditions to be screened	1. Conditions that women are interested to know	Condition to be screened	1.as many as possible
	2. High prevalent genetic conditions (not rare)		2. Severe conditions that being able to be confirmed by other tests. Treatments or interventions are available		2. non-rare diseases
Information from test	1. As much as possible on this risk/probability that baby might have genetic problems	Information from test	1. Reliable and comprehensive information; Results are easily interpreted	Information from test result	1. Information on risk of disability
	2. Simple information		2. Simple information. Difficult to make prognosis		2. Medical implications
Test performance	1. Not consider a test with false positive rate higher than 5%	Test performance	1. Low false positive and false negative rates	Test sensibility	1. Equal or less than 75%
	2. Detection rate: 50% - 95%		2. Higher false positive rate to maximize number of cases		2. 76% - 85%
	3. Detection rate: above 95%			Test specificity	3. 86-95%
	4. Take the test regardless detection rate to reduce risk of any possible problems to the baby				4. Equal or above 96%
					1. Equal or less than 75%
					2. 76% - 85%
					3. 86-95%
					4. Equal or above 96%

Pregnant Women result	1. As soon as possible	Decision-Makers	Consensus results	1. Before the third regular pregnancy visit
	8 – 10 weeks			2. Before the second (10-12 week) regular pregnancy visit
	2. After 10 weeks	Test procedure	1. Test procedure and confirmation tests are simple and rapid which are not resulting in stresses on women	1. simple
			2. Complex test procedure causing stress on women	2. complex
Cost related to the test	1. \$0 (Cost-free access)	Cost related to the test	1. Affordable cost accepted by pregnant women	1. 0\$
	2. \$100 - \$500			2. 100\$
	3. Willing to pay to have information of test regardless the amount of money		2. Reasonable cost for public system, not overcome threshold	3. 200\$
				4. 300\$
Test uncertainty	1. Not consider a test with low accuracy rate of severity	Test uncertainty	1. Not likely to accept low level of certainty regardless health outcomes: physical or intellectual	1. Regardless the uncertainty
	2. Take the test regardless the uncertainty level (neither physical nor intellectual problems)		2. Accept uncertainty but depending on severity of screened conditions	2. Accept low accuracy rate on physical problems
	3. Prefer a test with higher accuracy rate of intellectual problems			3. Not taking test with the uncertainties

problems		
Pregnant Women	Decision-Makers	Consensus

Table 3 displays the initial list of ten attributes based on the identification of categories and decision points. Seven attributes ('condition to be screen', 'information provided from test result', 'cost of test', 'test uncertainty', 'test performance', 'test sensibility' and 'test specificity') were built from what was said by both groups of participants. Two attributes were built from what was said by women only ('moment at gestational age to obtain the test result', 'geographical access to the test') and one from what was said by decision-makers only ('test procedure'). Although based on responses provided by a single group, these attributes were retained by the researchers on the basis that they could be of interest to the other group. Yet, this hypothesis would be tested in the next step of the construction of the instrument.

Table 3
Potential attributes and levels for consultations and focused group discussion

Attributes	Levels
1. Conditions to be screened	<i>1. can be treated or lead to a termination of pregnancy</i>
	<i>2. can be treated or lead to termination of pregnancy and should not be rare</i>
2. The test result that is presented to a pregnant woman is about	<i>1. the risk of disability</i>
	<i>2. the risk of disability and its medical implications</i>
	<i>3. the risk of disability, its medical and social implications</i>
3. Test performance	<i>1. known</i>
	<i>2. uncertain</i>
4. Test sensibility	<i>1. less than or equal 75%</i>
	<i>2. 76 to 85%</i>
	<i>3. 86 to 95%</i>
	<i>4. equal or above 96%</i>
5. Test specificity	<i>1. less than or equal 75%</i>
	<i>2. 76 to 85%</i>
	<i>3. 86 to 95%</i>
	<i>4. equal or above 95%</i>
6. Moment at gestational age to obtain the test result	<i>1. before the third prenatal visit</i>
	<i>2. before the second prenatal visit</i>
7. Cost related to the test	<i>1. 0 \$</i>
	<i>2. 100 \$</i>
	<i>3. 200 \$</i>
	<i>4. 300 \$</i>
8. Degree of test result certainty to the severity of the disability	<i>1. the child will have a physical and/or intellectual disability</i>
	<i>2. the child could have the disease, but without it resulting in a physical and/or intellectual disability</i>
9. Test procedure	<i>1. simple</i>
	<i>2. complex</i>
10. Geographical access to the test	<i>1. in a local health care facility (e.g., CLSC)</i>

Attributes	Levels
	<i>2. the confirmatory test is done in a regional hospital</i>
	<i>3. the confirmatory test can only be done in a hospital located in a city center</i>

Attribute selection and framing were based on consultations with two decision-makers and a pregnant woman, and a focus group with one decision-maker (who participated in the in-depth interview and who agreed to participate in future studies) and three pregnant women (who had not participated in the first interviews). The consultations were done by email. Participants to the consultation process proposed to eliminate the two attributes mentioned only by decision-makers, *test specificity* and *test sensibility*, since the medical terms might not give much significant meaning to pregnant women and were difficult to understand by a layperson. This was confirmed by the focus group discussion.

The consultations and the focus group discussion also led to the rewording of some attributes and levels in order to facilitate the comprehension of the questionnaire (for example, 'test procedure' became 'test sufficiency'). The process reduced the questionnaire to eight attributes. Five attributes had two levels and three attributes had three levels. Amongst the attributes, the levels of 'cost related to the test' was built based on what women said about how much out-of-pocket money they were ready to give for the test, as well as the real cost of the NIPS test presently offered in the public health care system. Details are provided in Table 4.

Table 4
List of attribute and attribute levels for the pilot project

Attributes	Levels	Explication
1. Conditions to be screened	<i>1. can be treated or lead to a termination of pregnancy</i>	A test can detect as many conditions as possible, provided that in case of a positive result, medical intervention is then possible.
	<i>2. can be treated or lead to termination of pregnancy and should not be rare</i>	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 (degree of accuracy of the test result)	<i>1. known</i>	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 confirms or reject the first test result.
	<i>2. uncertain</i>	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	<i>1. before or at the third prenatal visit</i>	The result is communicated at the latest during the third prenatal visit, around the 24th week of pregnancy
	<i>2. before or at the second prenatal visit</i>	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	<i>1. the child is certain to have a severe physical and/or intellectual disability that will affect the child's quality of life</i>	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
	<i>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</i>	The result can detect an intellectual or physical problem but does not indicate the severity of the disability.
5. Test sufficiency	<i>1. a positive result can be confirmed during regular prenatal visits</i>	Screening interventions are offered to all women during a regular pregnancy visit
	<i>2. a positive result may require confirmation by tests that are not offered during regular visits</i>	Screening interventions may require additional interventions, such as additional visits or specific tests like amniocentesis

Attributes	Levels	Explication
6. 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. <i>the risk of disability and its medical implications</i>	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. <i>the risk of disability, its medical and social implications</i>	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. Geographical access to the test	1. <i>all screening services are provided at a local health facility (e.g., CLSC)</i>	Screening can be done at a facility near the pregnant woman's residence
	2. <i>if the test is positive, confirmatory testing is done at a regional hospital</i>	Full testing (screening and confirmatory testing) may require a pregnant woman to go to the local referral hospital
	3. <i>if the test is positive, confirmatory testing can only be done in a hospital located in a big city</i>	Full testing (screening and confirmatory testing) may require a pregnant woman to go to a hospital in an urban center
8. Cost related to the test	1. 0 \$	
	2. 150 \$	
	3. 300 \$	

Stage 2 and 3: Experimental design, construction of tasks and the DCE survey design

Figure 2 illustrates an example of a choice task in the DCE survey. An example of the full survey is available on request.

Stage 4: Results of the pilot-test

The pilot project conducted with 33 pregnant women (aged from 19 to 37 years) was completed within 10 weeks (from 5th July to 12th September 2021). Over a total of 115 invitations sent, 68 women agreed to participate in the study. Among which, 36 full responses were received (53% response rate).

The pilot project revealed that it took an average of 11 minutes for participants to complete the questionnaire (2 minutes in minimum; 49 minutes in maximum).

Table 5 shows that the 'test performance' attribute was the most contributive to the choices made, followed by 'moment in gestational age to obtain test result', 'test sufficiency', 'costs', 'test uncertainty', and 'information provided from test result' (p-values < 0.05). Two other attributes, 'condition to be

screened' and 'geographical access to the test', are the least influential to the choice made (p-values at 0.2218 and 0.3247 respectively). Furthermore, the analysis also revealed which attribute levels were preferred by pregnant women. These results allowed us to form a choice task containing a dominant option for the full-scale DCE study that will be used to evaluate if answers given are random or well-thought.

Table 5
Conditional analysis of effects

Attributes	Levels	DF	Coefficient	Standard Error	Wald Chi-Square	Pr > ChiSq	Entry order in the stepwise selection
Conditions to be screened	1	1	-0.3354	0.2745	1.4927	0.2218	7
	2	0	0	.	.	.	
Test performance (degree of accuracy of the test result)	1	0	0	.	.	.	1
	2	1	-1.3176	0.2776	22.5266	<.0001	
Moment at gestational age to obtain the test result	1	1	-0.7890	0.2674	8.7046	0.0032	2
	2	0	0	.	.	.	
Degree of test result certainty to the severity of the disability	1	0	0	.	.	.	5
	2	1	-0.8860	0.2740	10.4550	0.0012	
Test sufficiency	1	0	0	.	.	.	3
	2	1	-0.6799	0.2586	6.9098	0.0086	
The test result that is presented to a pregnant woman is about	1	1	-1.0263	0.3525	8.4742	0.0036	6
	2	1	-0.3915	0.3375	1.3459	0.2460	
	3	0	0	.	.	.	
Geographical access to the test	1	0	0	.	.	.	8
	2	1	-0.856	0.3198	0.0716	0.7891	
	3	1	0.3854	0.3291	1.3717	0.2451	
Cost related to the test	1	0	0	.	.	.	4
	2	1	-0.4190	0.3430	1.4927	0.2218	

The conditional logit model found most attributes to be statistically significantly associated with the choice (p value < 0.05), except 'geographical access to the test' and 'conditions to be screened' attributes (p values at 0.3247 and 0.2218 respectively). However, because the predefined threshold for retaining an attribute was fixed at 25% (36), only the 'geographical access to the test' attribute was excluded.

Statistical analyses did not suggest bringing modifications to the attribute levels. However, researchers decided to extend the maximum of cost paid by users of the test of \$1000 with a \$200 increment (6 level attribute) to reflect the real amount that women might have to pay for detecting some anomalies not presently listed in a prenatal screening program, with the NIPS test.

Stage 5: Refinement and full-scale DCE survey

The pilot project results were used to finalize the questionnaire for the full-scale DCE survey.

This final DCE questionnaire contains seven attributes (5 attributes with 2 levels, one attribute with 3 levels and one attribute with 6 levels) (Table 6).

Table 6
Final list of attributes and levels for full-scale DCE

Attributes	Levels
1. Conditions to be screened	1. <i>can be treated or lead to a termination of pregnancy</i>
	2. <i>can be treated or lead to termination of pregnancy and should not be rare</i>
2. Test performance (degree of accuracy of the test result)	1. <i>known</i>
	2. <i>uncertain</i>
3. Moment at gestational age to obtain the test result	1. <i>before or at the third prenatal visit</i>
	2. <i>before or at the second prenatal visit</i>
4. Degree of test result certainty to the severity of the disability	1. <i>the child is certain to have a severe physical and/or intellectual disability that will affect the child's quality of life</i>
	2. <i>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</i>
5. Test sufficiency	1. <i>a positive result can be confirmed during regular prenatal visits</i>
	2. <i>a positive result may require confirmation by tests that are not offered during regular visits</i>
6. The test result that is presented to a pregnant woman is about	1. <i>the risk of disability</i>
	2. <i>the risk of disability and its medical implications</i>
	3. <i>the risk of disability, its medical and social implications</i>
7. Cost related to the test	1. <i>0 \$</i>
	2. <i>200 \$</i>
	3. <i>400 \$</i>
	4. <i>600 \$</i>
	5. <i>800 \$</i>
	6. <i>1000 \$</i>

4. Discussion

The work presented in this study concerns the construction of a DCE instrument addressing the issue of adding a test to a prenatal screening program, which can be administered to both patients and decision-makers. Having an instrument built on the preoccupation of both groups available allows researchers to compare the desirability levels for a new test from both sides. It allows the decision-making process to

include in the data used to support their decision, comparable information provided by groups that can have different preoccupations, but whose concerns, from a social perspective, are equally relevant.

The instrument shows similarities in its attributes with other DCE instruments built for use in studies on prenatal screening of the fetus (37–40). All instruments have attributes related to the level of information provided by the test results, to the time, in gestational age, to receive the results of a screening test, and to the test sufficiency, i.e., the impact of the test on the need of further invasive/non-invasive procedures to confirm a screening result. All instruments have a 'test performance' attribute, presented sometimes in different terms as 'detection rate', 'accuracy rate' (37–40). Unsurprisingly, this concern was also shared by decision-makers involved in the construction of our instrument but described in terms of 'false-negative' and 'false-positive' rates.

However, two dimensions that are present in our DCE instrument, are not found in other DCE instruments: one regarding which conditions that should be screened and one regarding which certainty level of the test result regarding the severity of a disability could be accepted. The reason for the discrepancy is probably due to the fact that other DCE instruments focus on the use of tests to screen for common chromosomal anomalies, for which the performance of the test is high. Our instrument targets potential chromosomal conditions that are not screened for but could be added to a screening program. The performance of the detection test for these conditions tends to be lower. These conditions tend also to lead to a wide range of phenotypes, hence of the severity of disability (37, 39, 41–44).

Moreover, the construction of the DCE instrument shows that a consensus can be reached on a final version, even if some attributes have been proposed by only one group. This probably reflects the respect and interest of all participants for preoccupation with some attributes that were of little concern to them before the study, but whose evocation has aroused an interest. To our knowledge, only one DCE instrument, in the field of pharmacy subsidy decisions, might have been applied to both patients and decision-makers (45). Yet, this paper contains some uncertainties whether both the public and health decision-maker/experts were effectively involved in the identification of the questionnaire attributes. This paper doesn't provide information regarding if and how a consensus on the DCE instrument attributes between both groups have been looked for (45, 46).

5. Conclusion

This study shows that it is possible to develop a DCE instrument to elicit values for an intervention that reflects the demand and the supply sides of the health care systems. By expanding the range of stakeholders involved in the valorization of an intervention, such an instrument might contribute to the efforts deployed to address the societal value of an innovation. Yet, a DCE study is resource-consuming and might give results that are difficult to explain to the target audience. The acceptability of this approach is therefore an issue. Research on these added values of DCE in the world of economic evaluation is still warranted.

Abbreviations

HEE: Health Economic Evaluations; CBA: Cost-Benefit Analyses; cfDNA: cell-free Fetal DNA; CHUL: Centre Hospitalier de l'Université Laval; CUA: Cost-Utility Analyses; DCE: Discrete Choice Experiment; HTA: Health Technology Assessment; INESSS: Institut National d'Excellence en Santé et Services Sociaux; ISPOR: International Society for Pharmacoeconomics and Outcomes Research; NIPS: Non-Invasive Prenatal Screening; QALYs: Quality-Adjusted Life-years; WTP: Willingness-To-Pay

Declarations

Ethics approval and consent to participate

Ethics approval for this study was obtained from *Comité d'éthique de la recherche du CHU de Québec-Université Laval* (project 2020-4877), and permission was granted to enroll participants at the CHUL hospital in Quebec City (Canada).

Consent to publication

Not applicable.

Availability of data and materials

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

HMN conceptualized the scope and aims of the study, developed the method, collected data, conducted analyses, and drafted the manuscript. CL collected and analyzed the data. MB developed questionnaires on online survey platform and provided technical aid on respondent recruitments. JRG and LN contributed to the ongoing development of the manuscript. BS collected and analyzed data of the systematic review of literature. DR supervised all stages of the study. All named authors have approved the manuscript.

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Figures

Figure 1

Key stages of the DCE development process

Section A: Choice of preferred options (1/7)

In this section, you have to make 7 choices. Each choice is for 2 options. The options are two test (test A and test B) that are described according to the elements presented in Section A. You must choose which of the two tests you prefer. You can also say that you have no preference for these two options.

* A1. What option for the test do you prefer to have (one choice only)?

	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 (degree of accuracy of the test result)	known	uncertain
Moment at gestational age to obtain the test result	before or at the second prenatal visit	before or at the third 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 can be confirmed during regular prenatal visits	a positive result may require confirmation by tests that are not offered during regular visits
The test result that is presented to a pregnant woman is about	the risk of disability	the risk of disability and its medical and social implications
Geographical access to the test	all screening services are provided at a local health facility (e.g., CLSC)	if the test is positive, confirmatory testing can only be done in a hospital located in a big city
Cost related to the test	300 \$	150 \$

Please select an answer below

☐ Test A☐ Test B☐ None

Figure 2

Sample of a choice task in the DCE pilot testing project

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