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Non-invasive prenatal screening for trisomy 21: What women want and are willing to pay

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ABSTRACT

Objective: To investigate the attitude among pregnant women regarding non-invasive prenatal testing (NIPT) for detecting trisomy 21 (T21) and to quantify their willingness to pay for NIPT.
Methods: A questionnaire was administered to pregnant women who received counselling for first-trimester screening (FTS) in two hospitals and nine midwife practices in the Netherlands.
Results: A total of 147 women completed the questionnaire, yielding a response rate of 43%. If NIPT for detecting T21 were available, 81% stated they would choose to have this test, and 57% of women who elected not to undergo FTS in their current pregnancy would perform NIPT if available. Willingness to pay for NIPT was correlated with age and income, but not education level. The price that participants were willing to pay for NIPT was similar to the current price for FTS.
Conclusion: The pregnant women in our study had a positive attitude regarding NIPT for T21, and more than half of the women who rejected prenatal screening would receive NIPT if available.
Practice implications: Due to the elimination of iatrogenic miscarriage, caregivers should be aware that informed decision-making can change with respect to prenatal screening with the introduction of NIPT.

1. Introduction

Non-invasive prenatal testing (NIPT) can use cell-free foetal DNA circulating in maternal blood to detect chromosomal trisomy, and NIPT was recently introduced into clinical practice. NIPT has both high sensitivity and high specificity [1].

In the Netherlands, first-trimester screening (FTS) is currently offered to all pregnant women as part of a national antenatal screening programme that is based on the "informed choice" principle, meaning that the individual's decision is voluntary and made with full understanding of the circumstances, including all expected benefits, burdens, risks and available alternatives. Invasive testing using chorion villus sampling (CVS) or amniocentesis is offered when the risk of trisomy is ≥1:200. In the Netherlands, approximately 25% of women elect to receive FTS, which is low compared to other countries, and women over the age of 36 have the right to request CVS and/or amniocentesis.

Decision-making regarding prenatal screening includes preparing for the next step, which is an invasive procedure in the event of increased risk of trisomy 21 (T21, or Down syndrome). At this stage, the decision requires balancing the probability of having a child with T21 against the risk of a procedure-related (iatrogenic) miscarriage. The most frequently cited reason for screening is to gain both knowledge regarding the health of the foetus and reassurance [2]. The principal reasons for declining screening include unfavourable characteristics of the screening test, ethical and/or religious objections, post-testing anxiety or uncertainty, and risks associated with invasive testing [2].

These arguments suggest that if a near 100% accurate, non-invasive test for foetal trisomy were available, women may make different choices regarding prenatal screening. Depending on cost and/or availability, NIPT may eventually replace current screening methods.

Although nearly everyone in the Netherlands has medical insurance, the cost of FTS (approximately €150) is only reimbursed for women ≥36 years of age. We therefore asked whether – and how much – women would be willing to pay for NIPT for T21 with risk-free diagnostic certainty. The price that women are willing to pay might also reflect how women value the test's risk-free diagnostic certainty.

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2. Materials and methods

Data were obtained from questionnaires that were completed by pregnant women. Information regarding prenatal screening for T21 was provided in accordance with current guidelines. The questionnaires were distributed by midwives and doctors following patient counselling for prenatal screening within the patient's first trimester. Questionnaires were distributed to all women in their first trimester, independent of their expressed interest regarding prenatal screening. The women were recruited from August 2011 through December 2011 from two hospitals and seven midwife practices in two regions (Leiden and Amsterdam) in the Netherlands. All questionnaires were treated anonymously (no name or address was listed on either the questionnaire or the envelope). The questionnaires were returned to one central hospital in pre-paid envelopes. In total, 340 women were invited to participate.

Background information regarding NIPT was provided, followed by questions designed to determine the participant's attitude towards NIPT. NIPT for T21 was described as a safe test with high (nearly 100%) diagnostic accuracy. The first part of the questionnaire addressed women's attitudes towards receiving information regarding prenatal screening and the reason(s) they might accept or decline prenatal screening in their current pregnancy. The participants were asked to indicate whether they would prefer NIPT replacing screening and/or invasive testing. Content analysis was used. The visual analogue scale (VAS) was used; the VAS is a graphic tool with a 100-mm horizontal line; the left end is labelled "very uncertain", and the right end is labelled "very certain". The participants were instructed to indicate the point on the scale that corresponds best with their feelings regarding the question [3].

Willingness to pay (WTP) was assessed using a payment card, consisting of a list of nine costs ranging from €50 to €500. For each amount, the women were asked to indicate whether they would be willing to pay this amount for non-invasive screening for trisomy 21. If they indicated a willingness to pay more than €500, they were asked to indicate the maximum amount they would be willing to pay [4–6].

The last part of the questionnaire included sociodemographic questions regarding age, education level, religious preference and household income. *Education level* was determined by asking respondents to indicate their highest completed level of education. *Religious preference* was determined by asking respondents to describe themselves as belonging to one of the following eight categories: no religion, Catholic, Protestant, other Christian, Islamic, Hindu, Humanist, or Other (specify). Income was determined by asking the respondents to indicate the range corresponding with their monthly net household income.

The following hypotheses regarding the relationship between the aforementioned sociodemographic factors and WTP were tested:

- Higher-income respondents have a higher WTP.
- Highly educated respondents have a higher WTP.
- Older participants have a higher WTP.
- Religious participants have a lower WTP.

The questionnaires were developed and pre-tested in both healthcare workers and pregnant women ($n = 10$/group) to determine the clarity of information, and several questions and answers were then optimised based on this pre-test.

The Dutch legislation does not require informed consent for a prospective study using questionnaires if the results are handled anonymously. Data were analysed using SPSS 17.0.

Table 1
Sociodemographic characteristics.

	Participants ($n = 147$)	%
Age		
Mean age (range)	32.9 (21–44)	
SD	4.6	
Low (<36 years)	101	68.7
High (≥36 years)	46	31.3
Level of education		
Low	13	8.8
Medium	15	10.2
High	57	38.8
Academic	62	42.2
Religious affiliation		
Religious	46	31.3
Not religious	100	68.0
Missing	1	0.7
Income household per month (euro)		
<1500	2	1.4
1500–3000	28	19.0
>3000	101	68.7
Missing	16	10.9

2.1. Participants

Table 1 shows the demographics of the participants. The mean age of the participants was 32.9 years, which is older than the average age of pregnant women in the Netherlands (31 years) [7]. The percentage of women <36 (68.7%) and ≥36 years of age (31.3%) was consistent with the age distribution of pregnant women in the Netherlands [8]. Relatively few participants had a low level of education and/or low income.

3. Results

In total, 340 women were given a questionnaire and invited to participate in the study, and 147 women (43%) completed and returned the questionnaire.

In total, 79 respondents (54%) opted for FTS in their current pregnancy, 7 respondents (5%) opted for an invasive procedure (all of whom were ≥36 years of age), and 61 respondents (42%) rejected prenatal screening, including 5 respondents who also declined information regarding the availability of prenatal screening.

Forty-eight respondents (33%) were recruited by the two hospitals, and the remaining 99 participants (67%) were recruited by their midwife.

The reasons stated (via an open-text field) for choosing screening were "we want to obtain knowledge regarding the baby's health" (41%); "'I have a higher risk for having a T21 baby because of my age" (24%); "we want reassurance" (5%); "if we receive a diagnosis of T21, we will terminate the pregnancy" (8%); "preparing for a possible child with Down syndrome" (4%); "if the child has T21, I do not want to burden my other children with the care of this child" (4%); and "I received screening during a previous pregnancy" (1%); 13% did not provide a reason.

The reasons for declining prenatal screening (indicating more than one reason was possible) included ($n = 61$) "not wanting to gain knowledge regarding T21 (15%); "I do not want to perform an invasive follow-up test" (23%); "I am opposed to terminating a pregnancy" (33%); "women felt that their risk of having a T21 child was too low to warrant testing" (41%); unfavourable features of the test (46%); and "I cannot or do not want to pay for FTS" (10%).

All 86 participants who opted for FTS in their current pregnancy expressed a positive attitude towards NIPT. Among the respondents who did not receive prenatal screening, 57% ($n = 35$) said that