



## ORIGINAL ARTICLE

# Implementing non-invasive prenatal testing (NIPT) in the Netherlands: An interview study exploring opinions about and experiences with societal pressure, reimbursement, and an expanding scope

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## Abstract

The noninvasive prenatal test (NIPT) as the first trimester prenatal screening (FTS) for trisomies 21, 18, and 13 is offered to all pregnant women in the Netherlands. NIPT using genome sequencing allows for an expansion of the scope of FTS and the introduction of NIPT gives rise to ethical and societal concerns about deliberated decision-making, pressure to engage in screening, and possible lack of equal access due to the financial contribution (€175) to NIPT. We explored the opinions and experiences of pregnant women, who were offered FTS, about these concerns, and the possibility of a broadened scope. Nineteen pregnant women representing a diversity of backgrounds were interviewed using a semi-structured interview guide. Eight women did not opt for prenatal screening while 11 did (NIPT = 4, combined test = 7). Women experienced a free choice to accept or decline prenatal screening, despite sometimes receiving advice from others. Prior to pretest counseling, some women had already deliberated about what an abnormal test result would mean to them. Others accepted or declined FTS without deliberation. The current Dutch policy of requiring a co-payment was acceptable to some, who believed that it functioned as a threshold to think carefully about FTS. Others were concerned that a financial threshold would lead to unequal access to screening. Finally, pregnant women found it difficult to formulate opinions on the scope of FTS, because of lack of knowledge. Life expectancy, severity, and treatability were considered important criteria for the inclusion of a condition in NIPT.

## KEYWORDS

Decision-making, deliberation, expanding scope, genetic counseling, NIPT, psychosocial, reimbursement, societal pressure

Robert-Jan H. Galjaard and Sam R. Riedijk are considered as joint senior authors.

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## 1 | INTRODUCTION

The noninvasive prenatal test (NIPT) provides an easy form of first trimester prenatal screening (FTS). In the Netherlands, NIPT screens for trisomies 21, 18, and 13 and is offered to all pregnant women, costing them €175. When using genome sequencing, NIPT allows for an expansion of the scope of FTS. The introduction of NIPT gives rise to ethical and societal concerns about deliberated decision-making, pressure to engage in screening, and possible lack of equal access due to the financial costs of NIPT. This study examines to what extent these concerns matter to pregnant women and explores their opinions and experiences concerning FTS.

In the Netherlands, all pregnant women can choose to have a screening test to determine their chance of fetal trisomies 21 (Down syndrome), 18 (Edwards syndrome), and 13 (Patau syndrome; Mersy et al., 2013). From 2007 until April 1, 2017 this screening was mainly conducted with the first trimester combined test (ftCT). If the ftCT determines an increased likelihood ( $>1:200$ ) of (one of) these common aneuploidies, pregnant women could choose either invasive prenatal genetic testing or refrained from further testing (Oepkes et al., 2016; Taylor, Chock, & Hudgins, 2014). On the April 1, 2014, the possibility to opt for the noninvasive prenatal test (NIPT) in a national implementation study 'trial by Dutch laboratories for evaluation of non-invasive prenatal testing' (TRIDENT-1) was added.

Since April 1, 2017, all pregnant women in the Netherlands have a choice between no first trimester screening (FTS), the ftCT, or NIPT within the TRIDENT-2 study. First (and second) trimester screening is mainly offered by primary care midwives, in a separate consultation with a funded duration of 30 min (Martin et al., 2015). At the moment all pregnant women in the Netherlands must pay out of pocket for the ftCT (€170), and NIPT also requires a €175 contribution. Second-trimester screening sonography scans are fully reimbursed (RIVM, 2018).

The introduction of NIPT provides easy accessible FTS. Using genome sequencing, NIPT allows for an expansion of the scope of FTS. Pregnant women opting for the ftCT still have a choice, in case of an increased risk, between NIPT or invasive prenatal genetic testing as the follow-up test. NIPT entails important benefits for pregnant women: first, it is more sensitive and specific as compared to the ftCT. The sensitivity of NIPT is 97% for Down syndrome, 90% for Edwards syndrome, and 90% for Patau syndrome, while the combined test has sensitivities of 85%, 77% and 65% (RIVM, 2019) respectively. Second, the use of NIPT will reduce the need for invasive procedures and the concurrent risks of miscarriage. However, the introduction of NIPT also raises some concerns.

First, it is feared that NIPT as a first-tier screening test may lead to routinization. The routinization argument is a container concept, which has been conceptually and empirically unraveled elsewhere (Kater-Kuipers, de Beaufort, Galjaard, & Bunnik, 2018). Routinization may refer to: (a) that NIPT may lead pregnant women to venture into first trimester prenatal screening less thoughtfully, (b) that in the absence of a risk of miscarriage, NIPT may lead to societal pressures to participate in prenatal screening and to stigmatization of those who

forego screening (Lewis, Silcock, & Chitty, 2013), and (c) because NIPT can be conducted early in the pregnancy, it may result in the trivialization of abortion (Farrell, Agatisa, & Nutter, 2014; Farrimond & Kelly, 2013). However, concerns about informed decision-making, pressure to test, and stigmatization lack empirical evidence, which questions their validity (Kater-Kuipers, Beaufort, et al., 2018).

Second, there are concerns about the influence of reimbursement policies on pregnant couples' views and uptake of prenatal screening. Pregnant couples might easily or thoughtlessly opt for reimbursed screening, whereas nonreimbursed screening may lead to unequal access (Health Council of the Netherlands, 2016). As said before, at the moment all pregnant women in the Netherlands must pay a contribution for the first trimester screening. In contrast, second trimester screening sonography scans are fully reimbursed (RIVM, 2018). The uptake of first trimester screening is around 45% whereas over 90% of pregnant women choose the fetal anomaly scan in the second trimester. The difference in reimbursement policies might be one of the reasons why the uptake of these tests is different, besides the fact that many women opt for an ultrasound to see their unborn child (Bakker, Birnie, Pajkrt, Bilardo, & Snijders, 2012).

Third, whole genome NIPT can detect a wide range of fetal chromosome abnormalities in addition to trisomies 21, 18, and 13 (Morain, Greene, & Mello, 2013). At the moment, pregnant women in the Netherlands can choose for a NIPT that only reveals trisomies 21, 18, and 13, or a NIPT that also reveals abnormalities in other chromosomes, indicated as secondary findings. However, in the Netherlands fetal sex and sex chromosomal abnormalities are not communicated, because the ministerial license does not allow analysis of the sex chromosomes (Oepkes et al., 2016). Expanding the scope of NIPT could be beneficial for pregnant couples, because more pathogenic abnormalities in the fetus can be detected (Tamminga et al., 2015). However, concerns on this expanding scope of NIPT have been voiced. Several studies have suggested that an expanded scope of NIPT may undermine informed decision-making because of the increased quantity and complexity of pretest information counselors have to offer (Dondorp et al., 2015). Moreover, people fear that with an expansion of the scope, prenatal screening is on a 'slippery-slope' towards screening for minor abnormalities and cosmetic traits. Different studies have shown that both professionals and pregnant women have difficulty deciding where to draw the line for an expanded NIPT (Tamminga et al., 2015; van Schendel et al., 2014). In practice, the expansion of NIPT has already started in many clinics in many developed countries, including the United States and the Netherlands (Oepkes et al., 2016; Wapner et al., 2015). An expanded NIPT includes other trisomies in addition to trisomies 21, 18, and 13 and also subchromosomal aberrations and microdeletions. Professionals indicate an urgent need for ethical guidance to determine an appropriate scope of NIPT (Tamminga et al., 2015). In this context, knowledge of women's preferences with regard to the scope of NIPT is indispensable.

The aim of the study at hand is to examine the ethical and societal concerns about routinization, societal pressure, reimbursement,

and an expanded scope of NIPT. Interviews with pregnant women regarding their views about NIPT, its characteristics, its (lack of) reimbursement, and its scope were conducted. Previous interview studies on attitudes of pregnant women and partners regarding NIPT mainly focussed on how pregnant couples view NIPT and its different aspects, but remain hypothetical on the aspects of that is, societal pressure and reimbursement (Lewis et al., 2013; van Schendel et al., 2014). Furthermore, most studies were conducted a couple of years before the introduction of NIPT as a first-tier screening test, making the results less applicable to present day pregnant couples. This study will give a more in-depth insight of the views and opinions of pregnant women who have made the decisions about whether or not to engage in such prenatal screening tests.

## 2 | METHODS

For this study a qualitative research design was used. Semi-structured individual interviews were held to explore the experiences and opinions of pregnant women regarding first trimester prenatal screening and in particular NIPT. Ten interviews were conducted before the availability of NIPT to all pregnant women in the Netherlands, whereas nine interviews were conducted after this implementation. All women provided written informed consent before participating in this study. The research ethics review committee (METC) of Erasmus MC, University Medical Centre Rotterdam, exempted this study (MEC-2016-399).

### 2.1 | Participants

Individual semi-structured interviews were conducted with 19 pregnant women from four midwifery practices between June 2016 and June 2017. After 19 interviews no new information was attained and therefore data saturation was reached, no further interviews were conducted. Women were recruited through four different midwifery practices across the country. The researchers deliberately sought to include women with different ethnic and religious backgrounds, educational levels, and socioeconomic status. However, women who signed up for the study were mostly Caucasian, highly educated women. Women were interviewed throughout all phases of their pregnancy. All 19 women were offered first trimester prenatal screening; 11 of them opted for prenatal screening (NIPT or ftCT), whereas eight did not. None of the pregnant women who chose for first trimester prenatal screening obtained high-risk results. Characteristics of the participants can be seen in Table 1.

### 2.2 | Procedure

Pregnant women were recruited and interviewed by two of the researchers (IMB and AKK). Nine interviews were held in person and the other 10 by telephone. The interviews were guided by an interview guide (see Supplementary Material), and if necessary follow-up questions were asked. The individual interviews lasted between 30

**TABLE 1** Characteristics of interviewed pregnant women

Mean age	31.5 years (range 20–45)
Mean gestational age at interview	23.1 weeks (range 9–40)
Nationality	
Dutch	18 (94.7%)
Other	1 (5.3%)
Screening	
No	8 (42.1%)
Yes, ftCT	7 (36.8%)
Yes, NIPT	4 (21.1%)
Education level <sup>a</sup>	
Highly educated	12 (63.2%)
Lower educated	7 (36.8%)
Religious	
Yes	5 (26.3%)
No	14 (73.7%)
Children	
Yes	10 (52.6%; mean = 1.8)
No	9 (47.4%)

<sup>a</sup>Education Level: Highly educated: College educated or higher.

and 60 min. The pregnant women received a €10 gift card for their participation.

An interview guide was developed in a multidisciplinary team of clinical geneticists, gynecologists, medical ethicists, and medical psychologists. The themes found to be relevant for the interviews were discussed and appropriate questions were formulated. The interview guide made sure that the interviews entailed a reflection on women's own choices with respect to screening, their views on the different screening modalities (ultrasound, combined test, NIPT), the appropriate scope of NIPT, and their experiences (if any) of societal pressure to undergo prenatal screening or to terminate an affected pregnancy. Furthermore, we included questions about the reimbursement policies for the various screening tests and asked the pregnant women what influence—if any—the reimbursement policy had on their choices for prenatal screening.

### 2.3 | Data analysis

All interviews were audio taped and transcribed verbatim by IMB afterwards. After transcription, the interviews were analyzed using Nvivo software. Data analysis was conducted using thematic analysis (Braun & Clarke, 2006). Responses in the interviews were coded independently by AKK and IMB. Afterward, these codes were compared and any discrepancies were discussed until consensus was reached. From these codes topics were extracted, and clustered into main topics and subtopics in order to identify important themes in the interviews. Representative quotes from the interviews were translated from Dutch to English and presented to illustrate the different themes.

### 3 | RESULTS

The four themes that were examined during the interviews were pregnant women's: (a) reasons for choosing first trimester prenatal screening or not (routinization), (b) experiences of pressure from the social environment and society, (c) thoughts and expectations about payment for prenatal screening, and (d) views on the possible expansion of the scope of prenatal screening. These four themes will be presented consecutively below.

#### 3.1 | Women's views regarding prenatal screening and NIPT

Women who participated in the interviews had various reasons to accept or reject prenatal screening. Some women did not opt for screening because they did not think about it at all, they believed they were too young and not at risk, or thought the test result of the ftCT is difficult to interpret, or a combination of these considerations. Others preferred a worry-free pregnancy above knowing the health status of their fetus, or would not take action after an abnormal test result. For some, abortion was not an option because of their religious beliefs or because they thought they would not be able to handle its psychological burden.

Well, at my age anyway, the chance is just a bit smaller [for Down syndrome]. Besides, I would not terminate my pregnancy if it [the unborn child] does have Down syndrome. They could also see it at the 20-week scan, so I can still prepare myself for it. (I9, age 20, no prenatal screening)

The pregnant women who opted for first trimester screening also gave various reasons. Some chose screening because they wanted information about the health of their child, because they wanted to have the possibility to end their pregnancy in case of an abnormal test result, or because they wanted to be able to prepare for the birth of a disabled child.

I just really wanted to know if it [the unborn child] was healthy. I really wanted that little piece of certainty, I really liked that. (I7, age 27, combined test)

The characteristics of the NIPT, such as its reliability and easiness compared to the ftCT, make testing more attractive to women. Ten women were interviewed before NIPT became available as a first-tier test in the Netherlands. Most of these women indicated that they would have opted for NIPT if it was available for them during their pregnancy. The interviewed women expected an increase in uptake with the introduction of NIPT, although women also thought that when pregnant women do not want to participate in prenatal screening they still will not opt for it.

You are going to find out whether your child is healthy or not [with the ftCT and NIPT], and many people do

not want to know that. (...) There might be somewhat more [women who opt for NIPT than with the ftCT], because it is easier and more accessible (...). That could be the case, but I think that it [the uptake of ftCT vs. NIPT] would not differ very much. (I17, age 39, combined test)

A few participants made their choice concerning prenatal screening before they received pretest counseling, based on information on the internet, or flyers, or peers' experiences. At the same time some women had a general concern that other pregnant women might not think through their choice for NIPT, that some accept the NIPT offer thoughtlessly, viewing it as part of standard procedure, without reading information leaflets or thinking about the information they received during counseling. As one woman indicated:

Because I think, they already take so much blood, why do you not add that [NIPT] to that [those tests]. (I3, age 32, no prenatal screening)

Therefore, pretest counseling for FTS should emphasize choice awareness among pregnant women. According to the interviewed women, good counseling should further include medical information about the test, such as its process, the reliability and explanation about trisomies 21, 18, and 13, and the possible next steps. A few participants also mentioned that it is important to discuss the emotional impact of screening, including knowing in advance what they want to do with the test result. However, other women indicated that they did not think about what to do with the test result before engaging in prenatal screening. They first wanted to wait and see what the test result would be.

#### 3.2 | Experiences of pressure from social environment and society

Women had different thoughts about and experiences with social and societal influence on their choice for first trimester prenatal screening. Most of the women indicated that their social environment did not influence their opinion about prenatal screening. However, some women indicated that their social environment did influence their choice. These pregnant women did not experience this influence as pressure: they stated that they could still make their own individual choice. Most women, furthermore, stated that the counseling by the midwife did not change their opinion, but more so strengthened it.

Yes I told her [the midwife] in advance [that I did not want to opt for screening]. But she said she wanted to explain everything about the screening to me, so she did. But that did not make me change my mind. (I15, age 35, no prenatal screening)

A few younger women (age range: 24–30) in our sample expected influence from family or friends on their choice when they would be older, because then they would be at higher risk and

family and friends would stimulate them to opt for screening. Some of these women also indicated that friends and family asked them the question why they opted for screening while they were young. Two women mentioned a certain influence toward testing from healthcare professionals and got the idea that testing is more self-evident to professionals.

*Nobody said [during the counseling session]: you can also do nothing. (I11, age 40, NIPT)*

A few women believed that society participation in prenatal screening is portrayed as being self-evident amongst others caused by media attention for the introduction of NIPT. It is presented as a very reliable test, and as an improvement of prenatal screening. It is expected that every woman would opt for it. Some women also had certain worries that being pregnant becomes medicalized, or that utilizing available tests becomes the social norm. Furthermore, concerning the termination of pregnancy, a few women had the opinion that there is certain societal pressure, in two directions: one woman's opinion was that terminating a pregnancy is more self-evident than to carry an affected pregnancy to term.

*You will be judged [by society] when you decide to keep a baby with a severe disorder while you had the possibilities to detect the disorder. (I12, age 33, NIPT)*

Another woman had the opinion that it is less acceptable to choose termination of pregnancy and stressed the importance of complete information in the counseling.

*I think that people are opposed to it and look at you and ask if you are sure to do it [terminating the pregnancy]. I think that people do not easily opt for it and also do not easily accept from others that they choose it. (...) Because people do not really know the consequences of having such a child [with a disability]. (I17, age 33, combined test)*

Most of the participants did not experience pressure from the society to test or not test. Most women experienced that there is sufficient freedom to refrain from screening, and most women believed that you are free to either carry an affected pregnancy to term or to choose termination of the pregnancy. The pregnant women also believed that in society there is not one major opinion on the termination of pregnancy; there are different opinions, influenced by, amongst others, culture and religious beliefs.

### 3.3 | Thoughts and expectations about payment for prenatal screening

Opinions on the role of payment for prenatal screening were quite diverse. Some women thought that having to pay for a test did not have any impact on their decision about prenatal screening. Others

thought that asking a fee might have impact on their personal choice for screening. They expected to be influenced by the price of the test, and probably would not opt for it if it were expensive. Some thought that if the tests were free of charge they would certainly opt for screening, whereas they would not take part if they were asked to pay.

*If I did not have to pay I would definitely do it [the combined test]. But the fact that I have to pay really makes me think it is a lot of money. I almost did not want to do it [the combined test]. (I2, age 29, combined test)*

A few women thought €175 is a lot of money for people with limited financial resources, while others thought that it is acceptable to ask that fee. Some women indicated that they think that a reimbursement of the test carries the message that it is a standard practice.

*But if it is free of charge, then it is more as if it is included in the total package [of tests during pregnancy], like the ultrasounds. You do not feel obliged, but it seems that it is included. (I8, age 26, combined test)*

Moreover, women thought that more pregnant women would opt for screening if it was free of charge and the uptake would increase. Therefore several women suggested that asking a fee might function as a threshold and makes pregnant women aware that it is an important choice they have to make.

*I do not know, if it is completely reimbursed it is accessible for everybody [NIPT]. I think that people would take the test more often. But on the other hand you do have to think about it very well, about the consequences. Maybe if it is too accessible people do not think about it good enough. So maybe asking money [for NIPT] could help. (I10, age 29, NIPT)*

A few women stated that it is your own choice to become pregnant and therefore you have to pay for a prenatal screening test yourself. Other women thought that prenatal screening should be free of charge in order to eliminate any threshold and make the test equally accessible for all women.

*People differ in their incomes and then [by asking a fee] you get involved in the rich versus poor argument. I believe that in healthcare income should not matter, especially not in the case of an unborn child (I18, age 33, no prenatal screening)*

### 3.4 | Expansion of the scope of prenatal screening

The discussion on the expanding scope of prenatal screening was often difficult to understand for women. When asked about their

preferences, benefits, and disadvantages of an expanded scope, women found it difficult to formulate their opinions because of lack of knowledge.

I do not dare to say something about that. I did not learn about what kind of abnormalities there could be, because I assume that it [the child] was just healthy. In case of an abnormal test result of course you are going to look at what it means. (I7, age 27, combined test)

Some women expressed reservations regarding the expansion of prenatal screening. One woman mentioned that pregnant women (and their partners) would not have worry-free pregnancies anymore if abnormalities were detected. Others thought that it is a step too far or felt it would be like playing God. Furthermore, a few women thought that society wants to exclude all possible abnormalities and feared that society tends to select perfect children and would not accept people with a disability anymore. Moreover, they feared that abortion for less severe abnormalities might also become accepted.

Just in general, I am opposed to everything being placed in a medical framework. That you can already know so many things in advance [before the baby is born]. The question is of course where this [expansion] will stop. (...) So I think I am just against it [the expansion] going on and on. (I18, age 30, no prenatal screening)

Other women thought that an expansion of the scope of prenatal screening is positive, because it provides certainty, or they were in favor of an expansion because it might prevent a long search for a diagnosis when a child is born with unexplained symptoms.

I would appreciate it when the test becomes expanded. I think it is something good because it just provides more certainty. You know, you are giving birth to a whole new life. (I7, age 27, combined test)

Especially the question on the kind of fetal abnormalities pregnant women want to know was difficult for women to answer, because of unfamiliarity with such abnormalities. In the interviews several categories of disorders were discussed such as early onset, late onset, and neurological disorders, based on categories as used in clinical genetics practice. Women who positively evaluate (a certain) expansion of the scope often indicated that disorders with limited or no life expectancy should be considered for inclusion in the test. Other considerations related to the question on which disorders should be included in the test, are the severity of the disorder, and the child's prospects of living an independent and happy life.

It is difficult. My idea would be that it [NIPT] should concern severely disabled children. Children who could never live independently, who need a lot of

medical care, where you ask yourself if they could be happy at all. (I11, age 40, NIPT)

However, women said that it is hard to say something regarding such a difficult and hypothetical situation of expecting a child with a severe disorder and regarding what they would do with such knowledge. They did not know what they would decide in the case of an abnormal test result. However, the different perceptions pregnant women have of, for example, Down syndrome suggest that women have different perceptions of severity and quality of life. Some women believed that Down syndrome is not sufficiently severe and they would not terminate the pregnancy for it. Others would terminate a pregnancy for Down syndrome because the child will always need care and might have many problems.

Another important argument was life expectancy, which is often a reason not to include late onset disorders in a screening test. They believed that one can have a joyful life until your 40s or 50s, without knowing about the disease, and maybe there will be new treatments discovered in the meantime. In contrast, a few women indicated that they probably would want to know late onset diseases because it enables you to prepare for your own future and the child's future. Some women believed that an expanded test might enforce striving for a perfect child and also mild disorders might be included in prenatal screening in the future. They were worried about where the expansion would stop.

## 4 | DISCUSSION

Pregnant women gave various reasons to accept or reject the first trimester prenatal screening. Women mainly chose for FTS to prepare for the birth of an affected child, or to terminate an affected pregnancy. Preferring a worry-free pregnancy or not wanting to take action after an abnormal test result was the main reason for declining FTS. In concordance with other studies, NIPT was preferred over the ftCT by most women because of its reliability (Chetty, Garabedian, & Norton, 2013; van Schendel et al., 2016). Some participating women would terminate a pregnancy in case of an abnormality; others would never consider a termination.

With regard to the influence of pretest prenatal counseling on the decision whether or not to participate in FTS, some pregnant women already made their choice about screening before visiting their obstetric caregiver, whereas others made this choice after counseling. Most women indicated that counseling for first trimester screening should both include information on the tests, the process, and the conditions screened for, as well as a discussion on the emotional impact of screening and the possibility of receiving an abnormal test result, which has been described before (Martin, Hutton, Spelten, Gitsels-van der Wal, & van Dulmen, 2014).

Pregnant women indicated that it is important that women think about what they would do with the results from prenatal screening beforehand, which is also underlined by healthcare professionals.



Deliberation, defined as the weighing and considering of what prospective parents consider to be a worthy life for their child and what a termination of pregnancy would mean to them, is seen as a key aspect of informed decision-making (Riedijk et al., 2014). However, a few interviewed women stated that they did not deliberate themselves, even not after pretest counseling, because they want to take the screening process step by step and would only start considering what they would do with an abnormal test result when they actually receive one. To our best knowledge this discrepancy is not found in previous studies, although one study did find that not wanting to think about what to do with a possible abnormal test result can be a reason for pregnant women to decline prenatal screening (Garcia, Timmermans, & van Leeuwen, 2008a). This discrepancy gives rise to the question what should be the focus of the prenatal screening counseling. Currently, the main focus of pretest counseling is providing information (Martin et al., 2014), but our results suggest that merely providing information is outdated: some women prioritize deliberation about their choice. Other studies also found that pregnant women want more than only information provision. They would like to have decision-making support or even advice from their midwives, whether or not to test (Martin et al., 2013). These results also show that women's personal information needs and preferences regarding deliberation differ. This requires personalized counseling in which the counselor addresses such personal needs. Previous research found that midwives feel more comfortable with providing information than with inquiring about the feelings and thoughts of the pregnant couple (Martin et al., 2014). Future research could focus on the best way to layout a pretest counseling session for first trimester prenatal screening, to balance information provision and deliberation support, and make midwives' task a little less complex. Also, a deliberation-focused approach to pretest counseling might need to be differentiated, as a subgroup of women want to take part in the first trimester prenatal screening without imagining what a detected abnormality might mean to them and deliberating what reproductive decision they would make in response. To respect the autonomy of these women, they should be allowed to access screening without partaking in deliberation.

Pregnant women feel like they are free to have their own opinion about the first trimester prenatal screening. They made different choices with regard to first trimester prenatal screening, but all felt that they could make these decisions independently, without pressure from others. Some of the women indicated that their surroundings influenced their choice, such as their partner, parents, friends, or family with (shared) beliefs or views of life, or their obstetric caregivers. These pregnant women did not experience this influence as pressure: they stated that they could still make their own individual choice. This phenomenon was described in the literature before, in a study in which it was examined whether prenatal screening programs allow pregnant women to make autonomous choices. The women in that study also stated that they were influenced by others during the decision-making process, such as their partners, their midwives, and society, but they made their own choices without pressure by others (Garcia, Timmermans, & van Leeuwen, 2008b; van der Steen et al.,

2018). These results suggest that the concern that NIPT will lead to a societal pressure to take part in screening and/or to terminate an affected pregnancy (Gekas et al., 2016), is contradicted in this study. This suggests that, for the women pretest counseling in the context of NIPT, emphasized freedom of choice.

So, pregnant women do not *personally* experience any pressure to (not) engage in prenatal screening, however, some of them did express the concerns that in society there are certain expectations with regard to participation in screening and either termination of an affected pregnancy, or carrying this pregnancy to term. Earlier research has also shown that pregnant women are worried that NIPT may lead to pressure to engage in screening; however, none of these studies described pregnant women experiencing this pressure themselves (Lewis et al., 2013; van Schendel et al., 2014).

Pregnant women differed in their opinions regarding the reimbursement of first trimester prenatal screening. These different views could be explained by differences in test choice, personal (financial) situation, and other aspects. Pregnant women did agree that a lack of reimbursement could result in unequal access to healthcare.

Furthermore, pregnant women agreed that reimbursing a screening test carries the message that the test is standard practice, as can be seen with the second trimester sonography scan, of which the uptake is over 90% in the Netherlands (Liefers, Cruisberg, & Atsma, 2017). Pregnant women believed that by reimbursing first trimester screening, the uptake will increase and women might venture into prenatal screening less thoughtfully. Some women indicated that asking a (small) fee made them think about their choice. They believed that it would also make other pregnant women aware that prenatal screening is a personal, important choice. However, they did feel that for some women even a small fee might be too much, therewith causing these women to forego participating in screening, even if they wanted to. In sum, women underlined the importance of informed choice, to which a small fee might be conducive, but that should not be at the expense of equal access. This again stresses the influence of how screening is organized on the women's choices, and demands that in the screening offer and pretest counseling the choice aspect is emphasized.

These results show that both scenarios, a reimbursed screening offer and a non-reimbursed screening offer, might challenge the nondirectiveness of the screening offer and the related counseling, whereby nondirective means 'withholding any normative judgment regarding the obtaining and application of genetic information' (Oduncu, 2002). Reimbursed first trimester prenatal screening might imply for pregnant women that the screening test is a good quality test, and participating in this test is self-evident and part of routine antenatal care. By contrast, a nonreimbursed screening offer might imply that the test offered is not seen as an important or of good quality by the healthcare providers, and therefore pregnant women would not want to opt for it. The effect of either message should be minimized in the counseling by explaining that while the test is reimbursed, women are still free to not opt for the test, or that while the test is not reimbursed, it is a good test that might provide options to

women. Adequate pretest counseling is the most important resource we have to counteract any negative effects of (not) reimbursing first trimester prenatal screening.

Finally, a possible expanding scope of NIPT turned out to be a difficult discussion point for pregnant women. Pregnant women found it difficult to make statements about the expansion of NIPT because they were unfamiliar with other disorders than the common trisomies currently included in first trimester screening. In the discussion, various categories were used, that is, early onset/late onset and actionable/non-actionable. Pregnant women were also not always familiar with these categories, in such cases examples to explain the categories were used, but the categories did make it easier for them to elaborate on the screening offer.

Some women were enthusiastic about an expansion because they thought that obtaining more information is something good. Others, however, were hesitant toward the expansion of NIPT and expressed the fear of a possible slippery slope. The interviewed pregnant women were made aware of the existence of a large number of serious conditions other than trisomies 21, 18, and 13 through this discussion, and some of them linked this to their own unborn child. Having this discussion could be burdensome for pregnant women, who could start questioning whether they are the ones having to decide on the screening offer.

Analysis of the responses given by the pregnant women on the questions regarding the scope of NIPT showed that they consider three things to be important in deciding whether or not to screen for a certain condition: (a) severity, (b) life expectancy, and (c) the possibility of an independent and happy life. If a condition would have (one of) these characteristics most women agreed that it should be included in the screening.

In the literature, to help women make individualized decisions about the scope of prenatal screening, it has been suggested that women should choose from a menu of options (Bunnik, de Jong, Nijssingh, & de Wert, 2013), with different categories of conditions included in the screening offer. Also, in another interview study pregnant women favored 'pure choice' model for expanded NIPT, wherein reproductive autonomy and informed choice are used to justify any prenatal screening decision a woman wants to make (Vanstone, Cernat, Nisker, & Schwartz, 2018). According to the findings of this study, such models would lead to practical problems: women had different interpretations of categories and found it hard to imagine what learning particular test results might mean to them and their child. Moreover, women had little knowledge of—or experience with—conditions that could potentially be included in the test, which raised the question whether women can make an informed, autonomous choice. From this the conclusion could be derived that the scope of NIPT should mainly be determined by experts, not by women themselves. Which experts should decide on the scope of NIPT should be determined by future research. Based on earlier research an expert panel in the Netherlands could include midwives, gynecologists, clinical geneticists, laboratory specialists, policy makers, and ethicists (de Jong et al., 2013; Kater-Kuipers, Bunnik, de Beaufort, & Galjaard, 2018). Nevertheless, opinions of women on

the scope of prenatal screening, as found in this study, are important inputs for the determination of the scope.

A second issue raised by the expanded scope and its related informed choice is that some interviewed women wanted to receive the test results first, and only after something of relevance has been found, they would wish to learn more detailed information on the condition detected. These findings suggest that in case of an expanded NIPT women might prefer a layered counseling wherein information in several stages can be provided to women in order to prevent information overload, as is proposed in a layered consent model for personal genetic tests (Bunnik, Janssens, & Schermer, 2013). Personal preferences regarding informational need and deliberation could therewith be taken into account.

#### 4.1 | Strengths and limitations

The strength of this study is that we included pregnant women from different regions in the Netherlands, aiming to include varying opinions within our sample. Furthermore, we included women with different test choices, to make sure that all choices (no prenatal screening, NIPT, and the ftCT) were well represented within our sample. Finally, we held these interviews right before and right after the introduction of NIPT as a first trimester screening test for all pregnant women in the Netherlands, making it a current and non-hypothetical matter for all interviewed pregnant women.

For this study women signed up themselves, which may have caused a bias in our pregnant population. Women who are willing to participate in an interview about prenatal screening, might have different characteristics and opinions compared to pregnant women willing not to be interviewed. This might explain why in our pregnant group more than half (11 out of 19) of the interviewed pregnant women opted for NIPT or the ftCT, whereas in the entire Dutch pregnant population less than half (45%) opts for first trimester screening. Therefore, the interpretation of these results must be performed with caution, as these might not be generalizable to the entire Dutch pregnant population. Furthermore, although we actively sought other target groups, mostly Dutch, highly educated, nonreligious women participated, which may also cause problems in the generalizability of the results. Also, some of the interviews were conducted before the availability of NIPT to all pregnant women in the Netherlands, whereas other interviews were conducted after its implementation, which may have elicited different opinions. Finally, in our pregnant group most women were highly educated (12 out of 19) of which most (9 out of 12) chose for either the combined test or NIPT. In the lower educated group (7 women) only two women chose for first trimester prenatal screening. Because we conducted a qualitative interview study, we are not able to draw conclusions or elaborate on the fact that more highly educated women opted for first trimester prenatal screening than lower educated women. However, this finding is in line with previous research which concluded that higher educated women opt for NIPT more often (Gil, Giunta, Macalli, Poon, & Nicolaides, 2015).



## 5 | CONCLUSION

Our study shows that there is a varying and broad range of opinions about first trimester prenatal screening, NIPT, pressure to test, the reimbursement of screening and the expanding scope among pregnant women in the Netherlands. Women feel that they have a free choice to opt for or decline prenatal screening, even though they sometimes receive advice from others for their decision. Adequate pretest counseling is important to maintain this experience of choice liberty now that NIPT has become part of the screening offer. However, counseling might need a shift in focus toward deliberation about what women want to know about the health of their child and what they want to do with the results, taking into account personal informational needs—which is already started in all Dutch training institutions and midwife practices. The significance of pretest counseling for first trimester screening continues to be a factor of great attention. However, our study clearly shows two important social issues that should be addressed in counseling. First of all, freedom of choice should be emphasized and second, possible messages deriving from either reimbursed or nonreimbursed screening should be minimized. Most women felt that not fully reimbursing screening could prevent the routinization of NIPT, but that this may also cause unequal access to healthcare. Furthermore, women had difficulty making statements about expanding the scope of NIPT, but agreed that determining the scope should mainly be based on severe, life-threatening disorders. Finally, our results suggest that the scope of NIPT should be determined by experts (in the Netherlands these could include midwives, gynecologists, clinical geneticists, laboratory specialists, policy makers, and ethicists), not by women themselves.

## AUTHOR CONTRIBUTIONS

I.M. Bakkeren, A. Kater-Kuipers, E.M. Bunnik, A.T.J.I. Go, A. Tibben, I.D. de Beaufort, R.J.H. Galjaard, and S.R. Riedijk declare that they have all contributed to the manuscript and have all approved the final version.

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## COMPLIANCE WITH ETHICAL STANDARDS

### Conflict of interest

I.M. Bakkeren, A. Kater-Kuipers, E.M. Bunnik, A.T.J.I. Go, A. Tibben, I.D. de Beaufort, R.J.H. Galjaard, and S.R. Riedijk declare that they have no conflict of interest.

## Human studies and informed consent

The research ethics review committee (METC) of Erasmus MC, University Medical Centre Rotterdam, exempted this study (MEC-2016-399). This study was in accordance with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from all 19 individual participants included in the study.

## Animal Studies

This article does not contain any studies with animals performed by any of the authors.

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## SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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