

ARTICLE



Non-invasive prenatal testing (NIPT) and pregnant women's views on good motherhood: a qualitative study

Elisa Garcia^{1,2}, Lidewij Henneman², Janneke T. Gitsels-van der Wal³, Linda Martin³, Isabel Koopmanschap², Mireille N. Bekker⁴ and Danielle R. M. Timmermans¹

© The Author(s), under exclusive licence to European Society of Human Genetics 2021

Women's views on responsible motherhood influence decision-making regarding participation in prenatal screening. Previous studies showed that the probabilistic nature of the first-trimester combined test and the potential requirement for subsequent invasive diagnostics serve as legitimate reasons for women to exclude prenatal screening from their moral responsibilities. These moral barriers might now be less relevant with the introduction of the non-invasive prenatal test (NIPT) resulting in women feeling a moral duty to use NIPT screening as part of responsible motherhood. This qualitative study explores the impact of NIPT on women's moral beliefs about the meaning of prenatal screening in relation to responsible motherhood. We performed semi-structured interviews with 29 pregnant women who were offered NIPT as a first-tier screening test within a Dutch nationwide study (TRIDENT-2). Results show that the inherent uncertainty about the fetus's health despite improved accuracy and the lack of treatment for a detected disorder, combined with the possibility to obtain information about actionable anomalies through the fetal anomaly scan, support women's perspectives that NIPT is not an obligation of responsible motherhood. Acceptance of NIPT is considered to be a free decision related to the information each woman needs to be a good mother for her child and her family. Women's views may change when NIPT has expanded to include treatable or preventable conditions.

European Journal of Human Genetics (2022) 30:669–675; <https://doi.org/10.1038/s41431-021-00945-3>

INTRODUCTION

Prenatal screening for fetal abnormalities aims to provide prospective parents with information about the presence of congenital defects that can help them to make autonomous reproductive choices [1]. In ethical literature, prenatal testing is often presented as a means of fulfilling the parental responsibility of striving for the wellbeing of the future child [2–4]. Pregnant women are widely considered to be responsible for their children's health. As good mothers, they are expected to make use of all available measures that can help to improve the health of their fetus and to avoid harm due to disability such as avoiding harmful substances and leading a healthy life. In this context of maternal responsibility for the health of the fetus, developments in prenatal testing have extended the moral norm of good motherhood to the obligation to acquire information about the health status of the fetus and to act upon that information for the benefit of the future child [2–4]. Using testing to ensure a positive pregnancy outcome might thereby be associated with responsible maternal behavior [2–8]. This view is supported by empirical studies exploring women's reasons for accepting or declining prenatal screening for fetal aneuploidy [9–11]. These studies have revealed that women's views on responsible motherhood play a role in their choice regarding the acceptance of prenatal screening

[12–14]. When deciding about testing, women aspire to do what a good mother is expected to do in order to ensure a good life for their child [13–15]. Women's considerations concerning the significance of screening for good motherhood refer to the utility of the information given by the test for guaranteed fetal health.

For many years, prenatal screening for aneuploidies was performed using the first-trimester combined test (FCT) followed by invasive diagnostic testing to confirm initial abnormal results [13, 14]. Empirical studies show that the inaccuracy of the FCT (i.e., false negative and false positive results) and the necessity to perform invasive testing with a small risk of miscarriage to confirm a positive result served as moral reasons for not considering testing as a responsibility belonging to good motherhood [3, 4, 9–15].

The introduction of the non-invasive prenatal test (NIPT) in the last decade [17] might remove these reported (moral) barriers for accepting screening. NIPT analyses fetal DNA present in maternal blood to screen for a few common chromosomal abnormalities. Compared to the FCT, NIPT offers a higher detection rate for Down syndrome (trisomy 21), Edwards syndrome (trisomy 18), and Patau syndrome (trisomy 13). The test also has lower rates of false positives, resulting in fewer referrals for confirmative invasive testing and hence less procedure-related fetal losses [16, 17]. Since the introduction of NIPT, ethical concerns about NIPT, possibly

¹Department of Public and Occupational Health, Amsterdam UMC, Vrije Universiteit Amsterdam, Amsterdam Public Health Research Institute, Amsterdam, The Netherlands.

²Department of Clinical Genetics, Section Community Genetics, Amsterdam UMC, Vrije Universiteit Amsterdam, Amsterdam Reproduction and Development Research Institute, Amsterdam, The Netherlands. ³Department of Midwifery Science, Amsterdam UMC, Vrije Universiteit Amsterdam, AVAG, Amsterdam Public Health Research Institute, Amsterdam, The Netherlands. ⁴Department of Obstetrics and Gynecology, Utrecht University Medical Center, Utrecht, The Netherlands. ✉email: e.garciagonzalez@amsterdamumc.nl

Received: 20 October 2020 Revised: 9 May 2021 Accepted: 3 August 2021

Published online: 17 August 2021

Table 1. Demographic characteristics of participants, $n = 29$ and their choices regarding screening.

	<i>n</i> (%)
Age in years	
22–30	17 (58.6)
31–35	9 (31.0)
>35	3 (10.3)
Country of origin	
The Netherlands	25 (86.2)
Other Western countries	1 (3.5)
Non-Western countries	3 (10.3)
Educational level ^a	
Low	0
Intermediate	9 (31.0)
High	20 (69.0)
Gestational age	
<16 weeks	3 (10.3)
16–25 weeks	20 (70.0)
>25 weeks	3 (10.3)
Not available	3 (10.3)
Number of children	
0	15 (51.7)
1	12 (41.0)
2	1 (3.5)
>3	1 (3.5)
Religion	
Christian	7 (24.1)
Islamic	1 (3.5)
Jewish	1 (3.5)
Without religion	20 (69.0)
Decision regarding screening for fetal aneuploidy	
FCT	0
NIPT	19 (66.5)
FCT and NIPT ^b	1 (3.5)
No screening	9 (31.0)
Decision regarding 20-week scan (FAS)	
FAS	29 (100)
No screening	0

Percentages may not add up to 100 due to rounding.

FAS fetal anomaly scan, FCT first-trimester combined test, NIPT non-invasive prenatal testing.

^aLow: primary school, lower level of secondary school, lower vocational training. Intermediate: higher level of secondary school, intermediate vocational training. High: higher vocational training, university.

^bOne participant wanted to have FCT and NIPT.

resulting in a new (normative) duty to accept screening, have been considered largely from a theoretical point of view [18–21]. Many authors believe that the accuracy of the results and the lack of physical risks will compel women to redefine the meaning of screening for good motherhood, causing them to perceive the use of NIPT as a moral duty that is part of their parental responsibility for the health of their child [21–23]. Empirical research has explored women's attitudes and decision-making concerning NIPT, including its ethical implications [24–30]. These studies reveal an increase in women's acceptance of prenatal testing with NIPT due to the lack of risks for the fetus [26, 30]. There is however

a lack of studies exploring the effect of NIPT on women's moral views regarding the significance of prenatal screening in relation to their duties toward their child. In this study, we aim to contribute to fulfilling this lack by exploring the impact of NIPT on women's moral beliefs concerning the meaning of NIPT in the context of the duties belonging to responsible motherhood. We performed semi-structured interviews with pregnant women who had been offered NIPT as a first-tier screening test since 2017 within the Dutch nationwide TRIDENT-2 study. This study, licensed by the Dutch Ministry of Health, aims to explore relevant aspects of the introduction of NIPT into the Dutch prenatal screening program [31]. The possibility to choose NIPT as a first-tier test is particularly relevant for exploring the impact of a risk-free and highly accurate test on pregnant women's moral views regarding prenatal screening.

METHODS

This paper describes a qualitative study including women who were offered NIPT within the TRIDENT-2 study.

Setting

In the Netherlands, prenatal screening for aneuploidies has been available for all pregnant women since 2007. Up until 2017, women who wanted to be informed about prenatal testing were offered the FCT followed by invasive diagnostic testing to confirm initial abnormal results. From April 2017, pregnant women in the Netherlands have been offered the option to choose FCT or NIPT as first-tier screening test for trisomy 21, 18, and 13 on a trial basis. Women selecting FCT can still opt for NIPT when an increased risk is identified [31, 32]. After a positive NIPT result, women are offered the choice to have invasive testing performed to confirm the diagnosis. Women are required to pay for some or all of the costs of the first-tier screening test themselves (~€170 for the FCT and €175 for NIPT in 2017). NIPT as a second-tier test is reimbursed by health insurance. Additionally, pregnant women are given a choice to have a fetal anomaly scan (FAS) for structural anomalies at ~20 weeks of gestation free of charge. Counseling for prenatal screening (FCT and NIPT) as well as the 20-week FAS is provided in the first trimester in a 30-min session with a certified obstetric counselor, most often a primary care midwife [29].

Participants

We interviewed 29 pregnant women who were offered NIPT in the TRIDENT-2 study between September 2017 and February 2018: 20 women who opted for NIPT screening (one woman requested FCT in addition to NIPT) and nine women who declined screening. Women were recruited from three midwifery practices in the region of North Holland (Amsterdam and surroundings) and through snowball sampling. All participants were intermediate to highly educated Dutch women of no religious faith. To avoid any interference in the decision-making process, interviews were performed after participants had made their decision about prenatal screening for fetal aneuploidy. Inclusion criteria were: aged 18 and older, the ability to give informed consent, and a good command of the Dutch language. Recruitment was stopped when data saturation was reached. Table 1 shows the participants' demographic characteristics and their choices regarding screening.

All participants were provided written and oral information about the aim of the study before signing a written consent form for study participation, audio-recording, and the use of the transcribed data. Participants could withdraw from the study at any time. Participants received a small gift for their time.

Interview guide and procedure

The interview questions were prepared based on the literature and discussions within the research team. The interview guide included questions about women's experiences with the offer of prenatal screening and their reasons for accepting or declining the FCT or NIPT. Explicit questions were included to assess whether women felt free in their choices and to explore the impact of other people (i.e., loved ones, medical professionals), and of social norms on their final decisions. The dialogue was extended to women's views concerning the moral significance of NIPT in relation to responsible motherhood (see Supplementary Table S1).

After performing the first three interviews, the wording of some questions was modified to make them easier to understand. All interviews were recorded and transcribed verbatim. All participating women were assigned anonymized identification numbers to protect their privacy.

EG and IK conducted the interviews. Women were interviewed at the location they indicated that they were assured of feeling comfortable. Most women were interviewed in person at their home ($n = 26$), two women at their workplace, and one woman by telephone. The woman's partner was present during five interviews. The duration of the interviews was between 45 and 90 min. To facilitate women to openly express their views and thoughts, we emphasized our neutral position and made sure that they understood that there were no right or wrong answers. Women were given the option not to answer those questions they did not want to. None of the women made use of this possibility.

Data analyses

Interview transcripts were analyzed using Atlas.ti 8 software. Data analysis was conducted using thematic analysis with a focus on ethical issues [33]. The initial analysis was aimed at identifying important data features that defined the nature of an answer to a research question. The resulting codes were grouped in clusters of similar meaning to determine relevant themes. After reviewing and, when necessary, redefining, the themes were clustered into main groups related to the moral significance of NIPT in relation to responsible motherhood.

Five interviews were independently coded by three researchers (LH, JTG, EG) to increase reliability. Codes were compared and discrepancies were discussed and resolved. All other interviews were analyzed by EG.

RESULTS

We identified four main themes relating to women's views regarding the significance of NIPT for morally responsible motherhood. Representative quotes are presented in Table 2.

The limited information from NIPT despite high accuracy

Although women who used NIPT and those who declined it valued the high accuracy of the test, all participants did not believe there was a moral duty to choose it. Participants reported that the lack of complete certainty about the health of the fetus was the main reason for not considering the use of NIPT as morally obligatory. They argued that NIPT only shows the probability of the fetus having one of the conditions screened for, without excluding the presence of other disabilities (Table 2, quote 1.1). The participants also reported that test results lack information about the severity of Down syndrome and the impact on the quality of life of the child as reasons for not considering NIPT morally mandatory (quote 1.2).

Another reason mentioned was the impossibility of avoiding many other potential complications during pregnancy or during delivery despite NIPT (quote 1.3).

Lack of preventive options other than pregnancy termination for the disorders targeted by NIPT

When asked about the relationship between responsible motherhood and the use of NIPT, women discussed the acceptability of pregnancy termination in the case of a confirmed disorder. All of them stated that morally responsible motherhood does not require screening for disorders that can only be prevented by terminating an affected pregnancy. Termination of pregnancy was described by some women as conflicting with their moral beliefs, as it implies rejecting being the mother of a disabled child (quote 2.1).

Differences were reported regarding the ethical acceptability of termination for the different disorders detected by NIPT. Participants considered termination in the case of Edwards syndrome and Patau syndrome as ethically acceptable due to the severity of these disorders (quote 2.2). Termination was described as a moral responsibility since these disorders are associated with low life expectancy. Screening for these disorders was also reported as being within the interests of the parents and

other family members to avoid the grief caused by a stillbirth or the birth of a child with a short lifespan (quote 2.3).

Most women believed that Down syndrome may be compatible with a satisfying life. Therefore, they considered the information provided by NIPT as insufficient for making a well-informed decision regarding the birth of a child affected by Down syndrome (quote 2.4). Although participants anticipated a significant impact on their lives, most women who chose NIPT reported that it was not their intention to avoid having a child with Down syndrome but used the test as preparation for the birth of a child with special needs (quote 2.5). Nevertheless, participants indicated that termination could be morally acceptable if prospective parents think that they cannot properly care for a disabled child (quote 2.6).

Presumed possibility to obtain information about treatable abnormalities through the fetal anomaly scan (FAS)

Participants expected FAS to give them reliable information about anomalies for which treatment after birth may be available. They stated that every woman should make use of FAS for obtaining information that could help them take measures for improving the health of the child or terminate the pregnancy in the case of severe anomalies (quote 3.1).

Women who used NIPT and those who declined it differed in their perception of NIPT versus FAS. Test acceptors described both tests as complementary, with the FAS as an additional screen for anomalies that cannot be detected by NIPT. Most of them expected extra confirmation that the baby was healthy (quote 3.2). For decliners, the availability of FAS made NIPT unnecessary. They saw the information from FAS as sufficient for gaining reassurance about the presence of anomalies, including Down syndrome (quote 3.3). Some decliners argued that the FAS would always be necessary for obtaining more information about the health of the child when NIPT indicates the presence of a disorder (quote 3.4).

Parental autonomy regarding participation in screening and the birth of a disabled child

The safety and high accuracy of NIPT were not mentioned as compelling reasons to take the test. Participants who made use of NIPT and those who did not have the test believed that it was up to the parents to decide whether they wanted to know in advance about the health status of the fetus and how to proceed once the results of the test were known (quote 4.1). Nevertheless, some participants speculated that societal pressure on expectant mothers toward the use of screening and termination of an affected pregnancy could increase because of the improved characteristic of NIPT (quote 4.2).

Though all participants stated that they made their own decision freely without being constrained by societal expectations, the support of their family for raising an affected child was reported as a requirement for continuing with an affected pregnancy. Women indicated that without help, they would be compelled to terminate the pregnancy even when they would have chosen to have the child (quote 4.3).

When discussing the impact of the availability of NIPT on reproductive autonomy, some women mentioned the costs of the test. While none of the participants reported the payment as a reason for not participating in screening, they were concerned that costs might undermine a free decision for parents who lack financial resources. At the same time, participants reported the cost factor as fostering informed decision-making, since it could prevent women from opting for screening just because it is offered free of charge (quote 4.4).

DISCUSSION

This qualitative study of women who were offered screening through FCT and NIPT in the Netherlands explores the potential

Table 2. Main themes relating to women's perspectives regarding the use of NIPT and responsible motherhood, illustrated with representative quotes.

Theme	Representative quote	Quote #
Limited information from NIPT despite high accuracy		
NIPT only shows the probability of the fetus having one of the conditions screened for without excluding the presence of other disabilities.	P016 (NIPT): Despite the leaflet and the counseling you don't know much, also about those syndromes. [...] There are so many more syndromes the child can have [...] and which can also be tested! Why these three?	#1.1
NIPT does not indicate the severity of DS.	P003 (NIPT): Suppose they say to you: "There is a high chance that your child has DS, thus we will do further research". And then you need to wait until the results of amniocentesis come back, to know whether it is yes or no. But they still do not know how severe it is!	#1.2
Impossibility to avoid many other potential complications in the course of a pregnancy or during delivery despite NIPT.	P001 (no test): You never know in advance how things will go. And that's naturally the case with pregnancy. You could do such a test; and it may appear that everything is okay but still a lot can happen in the course of the pregnancy or during the birth itself: the child may not have enough oxygen and be disabled for life.	#1.3
Lack of preventive options other than pregnancy termination for the disorders targeted by NIPT		
Termination of pregnancy as conflicting with responsible motherhood since it implies rejecting being the mother of a disabled child.	P005 (no test): The only extra option you get through the NIPT is the right to choose not to have the child. I don't think that's good motherhood. I consider that rejecting motherhood. P024 (NIPT): I think that if the child dies soon after birth or does not even make it. Look, if that were the case, I think it is easier to terminate the pregnancy, [...] That is really different from DS, I think. Because you can still lead a good life. At least the baby. So I really believe that. It depends on how bad it all is.	#2.1
NIPT as preparation for the birth of a child with special needs.	P025 (NIPT): It is not the case that I would terminate if my child has some disabilities. I want to look for available options. I thought that if I knew in advance and could prepare myself knowing that there is a big chance that my child will be unhealthy, then it would be easier for the child to come into the world.	#2.5
Termination of a DS pregnancy compatible with responsible motherhood if women think that they cannot take good care of the child.	P026 (no test): I think that some women really believe that they cannot offer a child with DS a good life and that is why they think that it might be better to terminate. I don't agree with that, but I don't think this makes them bad mothers. So I don't have that judgment. I think that there are a few women who do that because they have something like that, I really don't like to have such a child. I don't think along those lines.	#2.6
Presumed possibility to obtain information about treatable abnormalities through the Fetal Anomaly Scan (FAS)		
Women should make use of FAS for getting information that could help them to take measures for improving the health of the child or to terminate pregnancy in case of severe anomalies.	P007 (NIPT): I think that it [FAS] is an obligation towards your child. Because you can see more things; maybe less serious things but things you can act upon. Or when you see that the child is so disabled that there is no chance of life, then you can terminate while you would continue a pregnancy if you hadn't known it	#3.1
FAS an additional test to screen for anomalies that cannot be detected by NIPT.	P020 (NIPT): I find especially with the twenty-week ultrasound that there's just a lot of additional things you can see, or what you can be extra alert for. Or (...) Or things that are not life-threatening - but for which it might be practical that you deliver in the hospital where there's a cardiologist.	#3.2
Information from FAS as enough for getting reassurance about the presence of anomalies including DS.	P018 (no test): Of course, a 20-week ultrasound is different than the NIPT, but actually they are similar since you can see whether the child has a disability [...]. Doing the two tests, for us, was the same thing twice	#3.3
FAS made NIPT unnecessary since FAS is always necessary for getting more information about the health of the child when NIPT indicates the presence of a disorder.	P005 (no test): The 20-week ultrasound gives me information that NIPT does not and that can help me and the obstetrician or gynecologist to take better care of the child. Suppose that it comes from the NIPT that the child has DS, you still need the 20-week ultrasound to know more about it; therefore the NIPT adds nothing.	#3.4
Parental autonomy regarding participation in screening and the birth of a disabled child		
Right of prospective parents to make their own choice regarding testing and the birth of a disabled child.	P012 (NIPT): Prenatal tests are not about good motherhood; It is about information for parents. I don't think you can oblige parents to get information about their child. If they welcome their child also when it has a syndrome, you cannot oblige them to obtain that information.	#4.1
Expected societal pressure for NIPT use.	P011 (no test): I think there will be women who experience pressure from their social environment, or from society, that they [disabled children] should not be born	#4.2

Table 2 continued

Theme	Representative quote	Quote #
Support of family for raising an affected child and continuing with an affected pregnancy.	P007 (NIPT): If you seek for information not for termination, but you want to care for the child, then you should indeed take the responsibility to do that as well as possible. If I would live here alone with my husband with few family around me, I don't think that continuing pregnancy would be a wise choice, because we need support of others.	#4.3
Costs as undermining a free decision for parents who lack the financial resources/ as fostering informed decision-making.	P015 (no test): On the one hand it is good [costs of screening] because people then will think about the choice they make.[...] But on the other hand, if you have financial problems or if you don't have the money it would be sad if someone can't do it [screening] because of that.	#4.4

DS Down syndrome, FAS fetal anomaly scan, NIPT non-invasive prenatal testing.

impact of advantageous characteristics of NIPT on women's moral beliefs concerning the meaning of prenatal screening in relation to responsible motherhood. We show that although participants valued the improved characteristics of NIPT compared to FCT, they did not believe that pregnant women had a moral duty to make use of the test in order to be a good and responsible mother. As was the case before the introduction of NIPT, participants believe that the use of prenatal screening should be left to the responsibility of each woman (and her partner) [12, 13]. This includes using the test results to make decisions about the birth of a child with a condition and/or for preparing themselves and their surroundings for the care of an affected child. Our results contradict the expectations posed by some authors that the availability of NIPT will place women's responsibilities toward their unborn child in a new perspective [18, 19, 21–23]. According to these authors, the availability of a risk-free, accurate screening test will have an impact on the normative meaning of prenatal testing, causing women to perceive the use NIPT as a moral duty of good motherhood [22, 23].

Our results suggest that participants were not opposed to obtaining knowledge about the health of the fetus. On the contrary, they felt a moral responsibility to obtain information that could be used to improve the health of their child. In particular, performing FAS is perceived as a duty toward their child. Participants' views about the non-normative character of NIPT appears to be related to the inherent uncertainty about the health of the fetus despite improved test accuracy, and the lack of treatment for the disorders targeted by the test. Since NIPT screens for a limited number of conditions, a normal NIPT result does not exclude the presence of other disabilities or complications, either during pregnancy, at birth, or later in life. Additionally, whereas NIPT enables to establish the presence of aneuploidies with a high degree of accuracy, it cannot predict the severity or nature of a detected condition. The inconclusive information about the health of the fetus has as a consequence that participants do not consider the use of NIPT as part of their moral duty to ensure a healthy child.

The uncertainty about the health of the fetus is closely related to the untreatable character of the conditions targeted by NIPT. Since there is no treatment to avoid a detected disability, testing for aneuploidies is associated with the decision of whether to terminate an affected pregnancy [34–36]. Termination for fetal anomaly is a laden moral issue that is justified to prevent the child suffering severely [5, 8, 37]. As Edwards syndrome and Patau syndrome are severe disabilities associated with high miscarriage rates and a low survival rate amongst those born alive [38], the decision to terminate can be considered morally justifiable and in some cases morally responsible. In agreement with this view, most participants reported termination for Edwards syndrome and Patau syndrome as being for the benefit of the child. Nevertheless, they did not consider the use of NIPT for the detection of these

syndromes as morally required. An explanation for these contradicting views can be found in the low life expectancy of children with these aneuploidies. Therefore, participants might not perceive termination as part of their moral duty to prevent the suffering associated with a (long) life with a disability. Consequently, as participants' narratives suggest, compliance with NIPT is not seen as a moral duty toward the child. Indeed, termination for these two trisomies was reported as being for the benefit of themselves and other family members to avoid the emotional harms associated with giving birth to a deceased child or a child with a short life expectancy.

The diversity of phenotypes associated with Down syndrome and the possibility of the child having a good life [39] make a decision about termination for this condition ethically less evident on the grounds of a moral duty to prevent suffering than in the case of trisomies 13 and 18. The impossibility of knowing about the child's physical and cognitive future development is an additional factor of uncertainty for participants for not considering NIPT as morally required. In line with other studies exploring women's decisions concerning termination for Down syndrome [40, 41], participants' assessment of the normative character of screening and termination for this condition is based on their capacity to give their future child and other existing children the care they need to lead a fulfilling life.

At present, the participants' moral views concerning screening and termination for Down syndrome are reflected in the positive attitudes toward Down syndrome in the Netherlands [9, 14]. These societal views and the existence of special medical care for children with Down syndrome are guarantees for autonomous reproductive choices by pregnant women and their partners. The current views may change, however, when NIPT is seen as part of normal prenatal care. Studies have shown that technological developments in prenatal care can result in a societal expectation to make use of available technologies to avoid those conditions that can be detected before birth [2–8]. Broad implementation of NIPT might then result in a societal norm to screen for Down syndrome as part of the normative conduct of expectant mothers and, consequently, affect women's perceptions on responsible motherhood when confronted with Down syndrome [8, 12, 28, 31, 42]. Studies in other countries with a long tradition of prenatal screening have shown an increase in termination for Down syndrome [43]. This possibility is reflected by the participants' expressed fear of social pressure to prevent the birth of a child with this condition.

The participants' view of NIPT as not being morally obligatory might also be related to the fact that in the Netherlands prenatal screening is offered with emphasis on women's "right not to know" and not as standard care [9, 14]. Additionally, women who wish to make use of NIPT have to pay for it, while FAS has been offered free of charge to all pregnant women since 2007 [44]. This approach might convey the message that screening through FAS is necessary

—and hence morally required—while NIPT is an additional test that women can use if they are willing to pay for it [45]. Even if participants do not consider the use of FAS imperative, their narratives show that the use of FAS is perceived as responsible behavior. Full reimbursement of the cost of NIPT might result in an increase in women's sense of responsibility to use testing.

The differing moral evaluations of FAS and NIPT appear to be also related to women's perceptions of the treatable character of the anomalies detected by these tests. The possibility to detect treatable conditions, in combination with information about the severity of a disability, might cause women to perceive using FAS as part of their moral duty to improve the health of their child. Participants' perception of FAS as a moral duty suggests that women's views about NIPT might change if it is expanded to include the detection of (prenatally) treatable or actionable conditions. Although invasive testing would still be needed to confirm the presence of a disorder, the introduction of NIPT for treatable conditions might lead to a shift in the moral significance of NIPT from a means for providing couples with opportunities for reproductive choice to a way to encourage the health of the child [46].

However, uncertainty about the health of the fetus will still remain since the positive predictive values for different genetic conditions is low and the clinical significance of some genetic defects will remain difficult to predict [47]. This gap between the expanded possibilities to prenatally detect a broad range of congenital conditions and the limits of technology to provide certainty about the health of the fetus gives prospective parents the space to make their own moral evaluation about the meaning of NIPT for good motherhood. Expansion of NIPT to a greater number of (treatable) conditions should ensure that the offer of the test does not result in a technological imperative that puts pressure on women to accept screening to show themselves as good mothers [22, 23]. As Gauthier argues, moral responsibility dictates that people make their own free decisions when confronted with moral challenges according to their values and beliefs [48]. In the context of prenatal screening, good motherhood should be a result of women's responsible moral deliberation about the perceived implications of their decision for their child. In order to foster autonomous and moral responsible decision-making, counseling should support pregnant women to reflect on their responsibilities toward their child in accordance with their views of good motherhood, rather than conformity with social expectations [49].

Strengths and limitations

A strength of the study is that both women who accepted screening and those who declined were included. A limitation of our study is that all participants were intermediate to highly educated Dutch women and most of them had no religious faith. As it is known that social and cultural characteristics such as educational and social status, access to social resources, and healthcare and religious observance are important factors in women's decisions concerning the acceptance of screening [50], further research with a more heterogeneous sample including women from different cultural groups and socioeconomic backgrounds is necessary to be able to draw more general conclusions about the real impact of NIPT on women's moral beliefs regarding the normative nature of prenatal testing. Moreover, although our prime intention was to reach both prospective mothers and fathers, in the end we decided to include only women due of the low number of male respondents. Consequently, the impact of the partner's views on women's ethical beliefs is lacking.

CONCLUSION

Based on our findings, we conclude that the inherent uncertainty about the health of the fetus, despite its improved accuracy, combined with the untreatable characters of the disorders

targeted by NIPT and the possibility to obtain information about actionable anomalies through the FAS, support participants' perspectives that the use of NIPT is not an obligation of good motherhood. Women in our study considered it beyond their parental duties to make use of a test that can only provide information about the presence of a limited number of conditions that can only be prevented through termination of pregnancy. Future research needs to explore how the inclusion of treatable or preventive conditions in NIPT may impact women's beliefs concerning the meaning of testing as part of their moral duties belonging to responsible motherhood.

This study was conducted in the Netherlands, where uptake of screening is relatively low compared to other European countries, partly reflecting the rather positive views on people with Down syndrome, and negative attitudes toward pregnancy termination for this condition [9, 14]. Social and cultural values and views toward children with a disability shape women's moral beliefs regarding the use of NIPT. Therefore, our findings should be interpreted in the light of the social and cultural values in the Netherlands. The moral evaluation of women in other countries may be different.

DATA AVAILABILITY

The interviews (datasets) generated during and/or analyzed during the current study are not publicly available due them containing information that could compromise research participant privacy/consent, but are available from the corresponding author on reasonable request.

REFERENCES

1. Wilkinson S. Prenatal screening, reproductive choice, and public health. *Bioethics*. 2015;29:26–35.
2. Rothman BK. *The tentative pregnancy: prenatal diagnosis and the future of motherhood*. New York: Viking; 1986.
3. Rapp R. *Testing women, testing the fetus: the social impact of amniocentesis in America*. New York: Routledge; 1999.
4. Tremain S. Reproductive freedom, self-regulation, and the government of impairment in utero. *Hypatia: J Fem Philos*. 2006;21:35–53.
5. Savulescu J. In defense of procreative beneficence. *J Med Ethics*. 2007;33:284–8.
6. Davis D. *Genetic dilemmas: reproductive technologies parental choices, and children's futures*. New York: Oxford University Press; 2001.
7. Rapp R. Refusing prenatal diagnosis: the meanings of bioscience in a multi-cultural world. *Sci Technol Human Values* 1998;23:45–70.
8. Shakespeare T. *Disability rights and wrongs*. Routledge, Oxon, Great Britain, 2006.
9. Crombag NM, Boeije H, Iedema-Kuiper R, Schielen PCJ, Visser GHA, Bensing JM. Reasons for accepting or declining Down syndrome screening in Dutch prospective mothers within the context of national policy and healthcare system characteristics: a qualitative study. *BMC Pregnancy Childbirth*. 2016;16:121.
10. Carroll FE, Al-Janabi H, Flynn T, Montgomery AA. Women and their partners' preferences for Down's syndrome screening tests: a discrete choice experiment. *Prenat Diagn*. 2013;33:449–56.
11. van Bruggen J, Henneman L, Timmermans DRM. Women's decision making regarding prenatal screening for fetal aneuploidy: a qualitative comparison between 2003 and 2016. *Midwifery*. 2018;64:93–100.
12. Garcia E, Timmermans DR, van Leeuwen E. Rethinking autonomy in the context of prenatal screening decision-making. *Prenat Diagn*. 2008;28:115–20.
13. Garcia E, Timmermans DR, van Leeuwen E. Parental duties and prenatal screening: does an offer of prenatal screening lead women to believe that they are morally compelled to test? *Midwifery*. 2012;28:e837–43.
14. Crombag NMTH, Vellinga YE, Kluijfhout SA, Bryant LD, Ward PA, Iedema-Kuiper R, et al. Explaining variation in Down's syndrome screening uptake: comparing the Netherlands with England and Denmark using documentary analysis and expert stakeholder interviews. *BMC Health Serv Res*. 2014;14:437.
15. Remennick L. The quest for the perfect baby: why do Israeli women seek prenatal genetic testing? *Socio Health Illn*. 2006;28:21–53.
16. Gil MM, Quezada MS, Revello R, Akolekar R, Nicolaidis KH. Analysis of cell-free DNA in maternal blood in screening for fetal aneuploidies: updated meta-analysis. *Ultrasound Obstet Gynecol*. 2015;45:249–66.
17. Fairbrother G, Burigo J, Sharon T, Song K. Prenatal screening for fetal aneuploidies with cell-free DNA in the general pregnancy population: a cost-effectiveness analysis. *J Matern Fetal Neonatal Med*. 2016;29:1160–4.

18. Gekas J, Langlois S, Ravitsky V, Audibert F, van den Berg DG, Haidar H, et al. Non-invasive prenatal testing for fetal chromosome abnormalities: review of clinical and ethical issues. *Appl Clin Genet*. 2016;9:15–26.
19. Dondorp W, de Wert G, Bombard Y, Bianchi DW, Bergmann C, Borry P, et al. Non-invasive prenatal testing for aneuploidy and beyond: challenges of responsible innovation in prenatal screening. *Eur J Hum Genet*. 2015;23:1438–50.
20. Vanstone M, King C, de Vrijer B, Nisker J. Non-invasive prenatal testing: ethics and policy considerations. *J Obstet Gynaecol Can*. 2014;36:515–26.
21. Schmitz D, Netzer C, Henn W. An offer you can't refuse? Ethical implications of non-invasive prenatal diagnosis. *Nat Rev Genet*. 2009;10:515.
22. Ravitsky V. The shifting landscape of prenatal testing: between reproductive autonomy and public health. *Hastings Cent Rep*. 2017;47:S34–40.
23. Ravitsky V. Non-invasive prenatal diagnosis: an ethical imperative. *Nat Rev Genet*. 2009;10:733.
24. Reinsch S, König A, Rehmann-Sutter C. Decision-making about non-invasive prenatal testing: women's moral reasoning in the absence of a risk of miscarriage in Germany. *New Gen Soc*. 2021;40:199–215.
25. Farrell RM, Mercer MB, Agatias PK, Smith MB, Philipson E. It's more than a blood test: patients' perspectives on noninvasive prenatal testing. *J Clin Med*. 2014;3:614–31.
26. Lewis C, Silcock C, Chitty L. Non-invasive prenatal testing for Down's syndrome: pregnant women's views and likely uptake. *Public Health Genom*. 2013;16:223–32.
27. Vanstone M, Cernat A, Nisker J, Schwartz L. Women's perspectives on the ethical implications of non-invasive prenatal testing: a qualitative analysis to inform health policy decisions. *BMC Med Ethics*. 2018;19:27.
28. van Schendel RV, Page-Christiaens GC, Beulen L, Bilardo CM, de Boer MA, Coumans ABC, et al. Trial by Dutch laboratories for evaluation of non-invasive prenatal testing. Part II—women's perspectives. *Prenat Diagn*. 2016;36:1091–8.
29. Bowman-Smart H, Savulescu J, Mand C, Gyngell C, Pertile MD, Lewis S, et al. Is it better not to know certain things?: views of women who have undergone non-invasive prenatal testing on its possible future applications. *J Med Ethics*. 2019;45:231–8.
30. van Schendel RV, Kleinveld JH, Dondorp WJ, Pajkrt E, Timmermans DR, Holtkamp KC, et al. Attitudes of pregnant women and male partners towards non-invasive prenatal testing and widening the scope of prenatal screening. *Eur J Hu Genet*. 2014;22:1345–50.
31. van der Meij KRM, Sistermans EA, Macville MVA, Stevens SJC, Bax CJ, Bekker MN, et al. TRIDENT-2: national implementation of genome-wide non-invasive prenatal testing as a first-tier screening test in the Netherlands. *Am J Hum Genet*. 2019;5:1091–101.
32. van Schendel RV, van El CG, Pajkrt E, Henneman L, Cornel MC. Implementing non-invasive prenatal testing for aneuploidy in a national healthcare system: global challenges and national solutions. *BMC Health Serv Res*. 2017;17:670.
33. Braun V, Clarke V. Using thematic analysis in psychology. *Qual Res Psychol*. 2006;3:77–101.
34. de Jong A, de Wert GM. Prenatal screening: an ethical agenda for the near future. *Bioethics*. 2015;29:46–55.
35. Shakespeare T, Hull RJ. Termination of pregnancy after non-invasive prenatal testing (NIPT): ethical considerations. *J Pr Ethics*. 2018;6:32–54.
36. McDougall R. Parental virtue: a new way of thinking about the morality of reproductive actions. *Bioethics*. 2007;21:181–90.
37. Savulescu J. Abortion, infanticide and allowing babies to die, 40 years on. *J Med Ethics*. 2005;39:257–9.
38. Irving C, Richmond S, Wren C, Longster C, Embleton ND. Changes in fetal prevalence and outcome for trisomies 13 and 18: a population based study over 23 years. *J Mater Fetal Neonatal Med*. 2011;24:137–41.
39. Antonarakis SE, Lyle R, Dermizakis ET, Reymond A, Deutsch S. Chromosome 21 and down syndrome: from genomics to pathophysiology. *Nat Rev Genet*. 2004;5:725–38.
40. Korenromp MJ, Page-Christiaens GCML, van den Bout J, Mulder EJH, Visser GHA. Maternal decision to terminate pregnancy in case of Down syndrome. *Am J Obstet Gynecol*. 2007;196:149.e1–11.
41. Lou S, Carstensen K, Petersen O, Palmhøj Nielsen C, Hvidman L, Retpen Lanther M, et al. Termination of pregnancy following a prenatal diagnosis of Down syndrome: a qualitative study of the decision-making process of pregnant couples. *Acta Obstet Gynecol Scand*. 2018;97:1228–36.
42. Skotko BG. With new prenatal testing, will babies with Down syndrome slowly disappear? *Arch Dis Child*. 2009;94:823–6.
43. de Graaf G, Buckley F, Skotko BG. Estimation of the number of people with Down syndrome in Europe. *Eur J Hum Genet*. 2021;29:402–10.
44. van El CG, Pieters T, Cornel M. Genetic screening and democracy: lessons from debating genetic screening criteria in the Netherlands. *J Community Genet*. 2012;3:79–89.
45. Bunnik EM, Kater-Kuipers A, Galjaard RJH, de Beaufort ID. Should pregnant women be charged for non-invasive prenatal screening? Implications for reproductive autonomy and equal access. *J Med Ethics*. 2020;46:194–8.
46. Dondorp WJ, Page-Christiaens GCM, de Wert GMWR. Genomic futures of prenatal screening: ethical reflection. *Clin Genet*. 2016;89:531–8.
47. Neufeld-Kaiser WA, Cheng EY, Liu YJ. Positive predictive value of non-invasive prenatal screening for foetal chromosome disorders using cell-free DNA in maternal serum: independent clinical experience of a tertiary referral center. *BMC Med*. 2015;13:129.
48. Gauthier C. Moral responsibility and respect for autonomy: Meeting the communitarian challenge. *Kennedy Inst Ethics J*. 2000;10:337–52.
49. Cernat A, De Freitas C, Majid U, Trivedi F, Higgins C, Vanstone M. Facilitating informed choice about non-invasive prenatal testing (NIPT): a systematic review and qualitative meta-synthesis of women's experiences. *BMC Pregnancy Childbirth*. 2019;19:27.
50. Gitsels-van der Wal JT, Verhoeven PS, Manniën J, Martin L, Reinders HS, Spelten E, et al. Factors affecting the uptake of prenatal screening tests for congenital anomalies; a multicentre prospective cohort study. *BMC Pregnancy Childbirth*. 2014;14:264.

ACKNOWLEDGEMENTS

We wish to thank all women and men who participated in this study. We thank the midwifery practices for their help with recruitment, and Elsbeth van Vliet-Lachotzki (VSOP Patient Alliance for Rare and Genetic Diseases) for her feedback on the interview guide and protocol. We thank all members of the Dutch NIPT Consortium (see Supplementary Appendix) for discussion and feedback on this study.

AUTHOR CONTRIBUTIONS

LH, DRMT contributed to the design and implementation of the research, to the analysis of the results and to the writing of the manuscript. They supervised the findings of this work. EG (first author) contributed to the design and implementation of the research. She developed the interview guide, performed some of the interviews and analysed all the interviews together with IK. IK contributed to the development of the interview guide, performed some of the interviews and contributed to the analysis of the results. JTGvdW contributed to the development of the interview guide and to the analysis of the results. LM and MNB contributed to the development of the interview guide. All authors discussed the results, provided critical feedback and contributed to the final manuscript.

FUNDING

Supported by a grant from the Netherlands Organization for Health Research and Development (ZonMw, No. 543002001).

COMPETING INTERESTS

The authors declare no competing interests.

ETHICS APPROVAL

Ethical approval for this interview study was obtained from the Medical Ethical Committee of the VU University Medical Center Amsterdam (no. 2017.165 (A2018.069)).

ADDITIONAL INFORMATION

Supplementary information The online version contains supplementary material available at <https://doi.org/10.1038/s41431-021-00945-3>.

Correspondence and requests for materials should be addressed to E.G.

Reprints and permission information is available at <http://www.nature.com/reprints>

Publisher's note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.