Ambivalence among women who gave birth after receiving a negative result on non-invasive prenatal testing: a qualitative study

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**DOI:** 10.21203/rs.2.12626/v1

**SUBJECT AREAS**  
*Maternal & Fetal Medicine*

**KEYWORDS**  
*ambivalence, genetic counseling, NIPT, anticipatory anxiety, content analysis*
Abstract

Background This study aimed to investigate the factors affecting ambivalent feelings among women who gave birth after having received negative results on non-invasive prenatal genetic testing (NIPT). Methods A questionnaire was sent to women who received a negative NIPT result, and a contents analysis was conducted for those 1562 women who responded to the open-ended question. The contents of these qualitative data were analyzed using the N-Vivo software package. Results Environmental factors, genetic counseling-related factors, and increased anticipatory anxiety affected the feeling of ambivalence among pregnant women. Furthermore, pregnant women desired more information regarding the detailed prognosis for individuals with Down syndrome and living with them and/or abortion, assuming the possibility that they were positive. Conclusions Three major interrelated factors affected the feeling of ambivalence in women. Highlighting and discussing such factors during the genetic counseling may resolve some of these ambivalences, thereby enhancing the quality of decisions made by pregnant women.

Background

The fact of having a prenatal test for fetal abnormalities may affect maternal anxiety [1, 2]. Pregnant women who made uninformed choice for non-invasive prenatal genetic testing (NIPT) feel more decisional regret associated with prolonged anxiety [3, 4]. Women typically have two conflicting thoughts at the early stage of pregnancy: the desire to know about the neonate via ultrasound examination and the reluctance to conceive negative news. Many women reportedly take the NIPT to relieve their
Ambivalence regarding negative results [5]. Furthermore, certain women feel like their pregnancy is provisional or temporary until they learn about their NIPT results, i.e., although they may feel physical changes, they may not acknowledge their pregnancy until they see their results [6]. Ambivalence can be defined as the “simultaneous existence of positive and negative evaluations of an attitude object” [7]. Ambivalence has been identified among the attitudes of the general public regarding prenatal testing; however, the origins of the ambivalence remain unclear [8, 9]. Lewis et al. reported that numerous women taking the NIPT have ambivalent feelings [10].

All pregnant women who took the NIPT were asked to participate on a questionnaire-based survey of the Japan NIPT Consortium conducted during the three sessions of post-test genetic counselling: (i) immediately after the test while showing the results and (ii) one year after the test. We previously analyzed the results of the survey taken immediately after the test [11]. This study aimed to investigate factors affecting ambivalent feelings in pregnant women after undergoing NIPT, focusing on their free responses to open-ended questions one year after the test, and to assess the types of genetic counseling that should be administered to reduce the amount of stress in these women after receiving negative results in the NIPT.

Methods

Design

The study is based on qualitative research methodology, by content analysis using the free-form description of the first year after questionnaire for women who received negative results of NIPT.

Research procedure
Pregnant women who are identified as being at an increased risk of chromosomal disorders due to advanced maternal age, the results of ultrasound marker or maternal serum marker tests, or a family history of chromosomal abnormalities or those who have a high risk of being a translocation carrier were eligible for NIPT. They underwent NIPT after taking genetic counseling by clinical geneticist and/or genetic counselor. We have reported the one-year results from a survey on awareness of genetic counseling [8]. After a year, a mail-in survey on women’s feedback regarding the NIPT was performed by the Japan NIPT consortium. The questionnaire was sent out to 7594 of 7740 women who received negative results in the NIPT between April 2013 and April 2014. The questionnaires, which were anonymous and self-administered, were collected at 34 centers. Of these 7594 women, 3251 (42.8%) responded. Contents analysis was conducted for 1562 respondents (20.6%) who included the free-form description to the open-ended question (Fig.1).

Participants
Because the first year after questionnaire was anonymous, and it was impossible to obtain the background data, the personal background of the respondents is unknown; however, the available basic background information of the respondents who underwent NIPT is shown in the Appendix (We have already reported the results of the awareness on genetic counseling of 7292 out of 7740 women[8], whereas this study was limited to those who received negative NIPT results among the 7740 women, and therefore, the background data were not exactly same)(Appendix Table.1and 2).

Data Analysis
The questionnaires were subsequently mailed to a third-party NIPT data center for
data entry. Qualitative methods are required in situations warranting detailed analysis and an understanding of the process to determine the nature of the issue being investigated. We undertook a combined content analysis (quantitative and qualitative) via text mining approaches, using N-Vivo Qualitative software for the free-form description part of the questionnaire. The advantage of computer-assisted content analysis of textual data is the coding reliability that helps to generate comparable results [12]. The text in the free-form description part was entered into the computer and used as input for the N-vivo software. Furthermore, all the text was read, and the coding process was discussed with two researchers. Thereafter, we reviewed the codes and grouped them into the primary categories with subcategories.

Ethics

To conduct this study, the participating centers obtained approval from their respective ethics committees

Results

We identified three primary categories generating feelings of ambivalence among women: (1) factors related to genetic counseling; (2) environmental factors, and (3) increased anticipatory anxiety. Furthermore, the former two factors caused the anticipatory anxiety (Table. 1).

Factors related to genetic counseling

This category [Factors related to genetic counseling] comprised two subcategories: [lack of information] (n = 159) and [Lack of psychological care adapted to individual needs] (n = 48). [Lack of information] further comprised two mid-level categories: “Inadequate support by medical staff up to taking NIPT” (n = 69) and “Lack of
information in case of positive result” (n = 91). "Inadequate support by medical staff up to taking NIPT” included (Lack of NIPT information from family physician before genetic counseling (26), (Family physician’s response) (n = 33), (Lack of neutrality in genetic counseling) (n = 11). “Lack of information in case of positive result” included (if the pregnancy was terminated) (n = 14), (if the pregnancy was continued) (n = 51), and (follow-up support to positive result) (n = 55). Lack of psychological care adapted to individual needs comprised “Lack of psychological care at genetic counseling” (n = 35), and “Lack of genetic counseling at the appropriate moment” (n = 17).

Regarding Lack of information, “support by medical staff up to taking NIPT” referred to the lack of support from the family physician during NIPT (Family physician’s response). Many family physicians were not only not helpful in referring their patient for genetic counseling, but they also themselves were unaware of it (Lack of NIPT information from family physician before genetic counseling). Owing to the limited number of NIPT facilities for pregnant women in Japan, some respondents also reported receiving a reproachful response from their family physician upon requesting information regarding NIPT or referrals (support from the family physician).

Furthermore, certain women were criticized for taking the NIPT during genetic counseling and many women wished that genetic counseling would be performed assuming that NIPT could give positive results regarding “Lack of information in case of positive result.” They wished to receive detailed information regarding the methods of terminating their pregnancy as one of the alternatives in case of chromosomal abnormalities in the fetus.

I don’t think it hurts to take some time to explain about the options for getting an
artificial termination of pregnancy in the case of a positive result. There is this idea that an abortion bad, but if the chance that you can love your child unconditionally is 0%, then I believe that it is an important option, even though it’s not easy to discuss...

(if the pregnancy was terminated)

However, some women who considered continuing their pregnancy despite receiving positive results upon NIPT were seeking information regarding the exact quality of life for individuals living with such illnesses and the social resources available for them.

What I really want to see added is what to do if the results do come out positive. I want to hear during the counseling session the exact types of social support that I could get if my child is born with Down syndrome or some other illness to help me live with and raise the child with the illness, for example at school, daycare, at home, names of organizations or municipal centers where I can get support.

(if the pregnancy was continued)

These data indicated that respondents were seeking information during genetic counseling that would support the decision to continue or terminate their pregnancy. These women desired information regarding the daily lives of children with disabilities and other related materials to guide their decision of whether they could raise a child with disabilities; furthermore, they desired follow-up support for women reporting with positive results in the NIPT.

Environmental factors

Environmental factors (n = 82) included the comfort levels of women for raising a child with disabilities; this aspect comprised three subcategories: Lack of awareness and education about diversity (n = 30), Insecurities on raising the
child\(n = 63\), and \(\text{Lack of social support systems for people with disabilities}\(n = 23\). \(\text{Insecurities on raising the child}\) comprised “Insecurities for the future” \((n = 26)\), “Vague insecurity” \((n = 20)\), “Insecurities for the child-rearing environment” \((n = 18)\), and “Familiarity with challenges of living with a disabled person” \((n = 9)\). “Insecurities for the future” included (After the parents’ death) \((n = 20)\) and (Impacts on the siblings) \((n = 17)\).

\(\text{The reason why many people have the image of prenatal testing as an “immoral thing” in Japan may come from views about ethics on this subject in Japan, or prejudice and closed-mindedness against people with Down syndrome and other chromosomal abnormalities. I just sometimes wish that those who are debating and giving their input on ethics would also take part in discussing how to change Japan into a more livable society for children with Down syndrome and their families.}\)

\(\text{(Lack of awareness and education about diversity)}\)

Regarding \(\text{Insecurities on raising the child}\), older parents had a sense of responsibility to raise the child; they were concerned regarding the life of the child after their death owing to their age at pregnancy, and they were concerned regarding burdening the siblings of the child and, thus, had “Insecurities for the future.”

Furthermore, certain respondents also exhibited “Vague insecurity” because raising a disabled child is unfathomable and difficult to imagine, owing to the lack of experience. Others, however, already had “Familiarity with challenges of living with a disabled person,” because they knew somebody or had family members with disabilities, thereby fostering the understanding of the merits and demerits and having discerned that it would be challenging. They also faced “Insecurities for the child-rearing environment” owing to their concern regarding an inadequate financial
support or the social environment for raising a child with disabilities.

As an actual mother of a child with chromosomal abnormalities, I’m glad that I had my baby, but raising my child is full of challenges and would be absolutely impossible without the cooperation of people around me, so it’s not something that can be glossed over. I got tested for my second pregnancy, but I don’t think I would’ve had the child if the results were positive (Familiarity with challenges of living with a person with disabilities).

Increased anticipatory anxiety

Increased anticipatory anxiety (n = 56) included the two subcategories Time-related anxiety (n = 47) and Anxiety about test accuracy (n = 9). Among the pregnant women who selected NIPT, there were some who were so anxious during the two weeks until they got the results that they could not sleep until finding out that their results were not positive. Also, knowing that test accuracy of NIPT was not 100%, some people were anxious that their result might be a false-negative, and they could not eliminate this anxiety until having the child.

Ambivalence

Ambivalent feeling comprised five subcategories: Options in the case of a positive result (n = 78), Guilt towards the child (n = 32), Criticisms on NIPT from others (n = 14), Denial of disabled people (n = 11), and How to tell the child (n = 4). Options in the case of a positive result included “Stated their decision” (n = 16) and “Difficulty stating their decision” (n = 62).

Some respondents “Stated their decision” with regard to the course of action in case of a positive NIPT result, whereas others expressed “Difficulty stating their decision” regarding continuing or terminating their pregnancy. In both cases, respondents seemed agonized over their decision in the case of positive results.
I had decided that if the results came out positive, I would not give birth to the baby. However, even though I now know that the results were negative, I haven’t told my own parents that I took the test. At the same time that I feel negative about the idea that we were picking and choosing who to keep if the results were positive, I also blame myself for my lack of confidence in raising the child, even though it is because I have health problems myself.

(Stated their decision)

Some women seemed to have a sense of Guilt towards the child after given birth and parenting the child, knowing that they were trying decide the life of the child.

I don’t think I would have taken the test if I were younger. Some people criticize that people take the test too casually because it places very little stress on the mother or child. but I believe that anybody who chose to take the test did so after putting lots of thought into it. Looking into my child’s face, sometimes I remember about when I took the test, and I can’t help feeling sorry towards my child. My baby’s life is very valuable, and I want to give all the love it deserves.

(Guilt towards the child)

Furthermore, some women could not tell anybody that they took the NIPT owing to social pressure Criticisms from others on NIPT, and others who felt guilty that having Denial of disabled people upon having children with disorders, which were screened during NIPT, despite obtaining negative NIPT results.

When I watch documentaries on TV, etc., and hear that the majority of opinions seem to support that “Whether or not a life should live should not be decided by parents, but should be treated as an independent life” “Even if my child has Down syndrome, I am so happy that they came to this world”. People who, for whatever
reason decided not to have the baby, will be burdened with guilt for the rest of their lives. I did test negative, but whenever I look into my child’s face and see a person with Down syndrome, I criticize myself because “I decided on her life.”

(Denial of disabled people)

Discussion and Conclusion

Discussion

The decision-making among pregnant women to receive a prenatal diagnosis is affected not only by individuals close to them, e.g., family members or friends, but also by the opinions of medical staff, the social environment, and uncertainty regarding fetal health conditions [12–15]. The results of this study also showed that factors related to genetic counseling and environmental factors greatly influence women’s decision-making regarding taking the NIPT. Because of the impact of factors related to adequate information, i.e., Factors related to genetic counseling[before undergoing NIPT and Environmental factors], which describe an inadequate social environment, the ability of independent decision making among women is perturbed, thereby further increasing the level of [anticipatory anxiety].

Furthermore, the guilt that women experience when deciding to give birth or abort the fetus, or the guilt they experience regarding decision-making related to disabled individuals and contrary sentiments associated with ideas that promote prenatal testing and that having to face this moral opposition induces a state of [Ambivalence] among some women even one year after taking the NIPT, whenever the unresolved feelings associated with the thought “What if the result had been positive?” resurfaced in their minds.

There are problems related to genetic counseling about assuming positive NIPT
results. Genetic counselors themselves are concerned that talking to parents about Down syndrome during prenatal testing is likely to elicit fear among them. Overly optimistic or negative information from medical staff, who largely influence the parents’ ultimate decision-making upon receiving positive NIPT results can also introduce a bias [16, 17], similar to the genetic counselors themselves who lack the necessary skills and knowledge [3]. Perhaps, this is focused on avoiding any negative effects of providing information while assuming a positive NIPT result on the mental status of pregnant women, who seek emotional relief from undergoing the test. However, many women also seek information regarding the precise living conditions of children with the screened genetic disorders and regarding the types of social resources available to them.

Statistics have revealed that in England, a few more women who received positive NIPT results chose to have an abortion compared to those who tested positive upon amniocentesis [3]. Concurrently, in Japan, the possibility of continuing the pregnancy existed among many women, despite obtaining positive NIPT results. The present results also show that it is important to offer genetic counseling under the assumption of a potentially positive NIPT result.

Among numerous pregnant women, the motivation to undergo prenatal testing stems from their need to “feel relieved [5].” However, among the women who inquire about prenatal diagnosis, many request information regarding not only about the actual test, but also regarding the many anxieties they may have about their children during pregnancy. Pregnant women attended NIPT genetic counseling sessions at a time when they could openly speak and be listened to; hence, it is very important to provide psychological support to pregnant women in addition to information regarding testing methods or disorders that can be screening during the
Fears regarding the potential discrimination faced by the child, prejudice, the lack of understanding among family members or other people, and the lack of societal support for raising the child are some of the reasons that motivate women to have a “healthy” child. Furthermore, the present results suggest that the society is not very open and livable for individuals with disabilities. Simultaneously, numerous individuals strongly felt the need to adapt to the social support systems for people giving birth to children with disabilities. These results suggest a positive and supportive perspective towards disabled individuals, fostering the hope that in the future, diversity will be widely accepted and that society will be welcoming to all children, irrespective of being born with disabilities. Many of the present study subjects were mothers with late-life pregnancies. We believe the potential impacts on family and siblings, societal prejudices on disabilities, and attachment towards the unborn child were weighed after careful consideration and understanding of societal prejudices and the increased chance of having a child with an abnormal chromosomal number owing to their age before choosing to undergo NIPT.

Similar to other types of prenatal testing, we believe that NIPT may also increase the pregnant women’s worries and anxieties about the unborn child. The rate of infertility treatment was high among the women who underwent NIPT (42.2%). Furthermore, women undergoing infertility treatment have strong fears regarding chromosomal abnormalities among their children [19, 20, 21]; however, informing women that results are usually negative for 98% of older mothers may as well reduce the anxiety among women undergoing infertility treatment [22, 23]. Offering detailed information to pregnant women during genetic counseling sessions to enable its juxtaposition with the mothers’ own experiences and values to make an
informed decision whether or not to undergo NIPT lowers the amount of uncertainty, stress, and anxiety they experience during the decision-making [24, 25]. The present results indicate that numerous individuals seek detailed information regarding the survival of children with positive results upon NIPT, including social support services, which indicates the lack of information and its accuracy regarding these disorders. As such, administration of NIPT is accompanied by high levels of stress among mothers, thereby potentially explaining their increased levels of anxiety until receiving the test results or their anxieties regarding the accuracy of the test [26].

Pregnant women taking the NIPT are compelled to gain adequate knowledge and understanding of the NIPT and the disorders screened therewith and to decide between continuing or terminating their pregnancy upon receiving a positive result within a very limited time. It is normal for all pregnant women to wish for a healthy child, which motivates them to take the NIPT to be “relieved [27].” However, even when they are relieved upon receiving negative results, many women were still ambivalent about their decision, feeling guilty about having tried to decide their child’s life, or having felt denial towards individuals with disabilities, but also having to endorse the thoughts about the test. This is probably what made them recognize their ambivalence and internal conflict at having these unresolved complex emotions.

Conclusion
The present study shows that even negative results can induce ambivalent feelings among pregnant women, and it is important to recognize that these feelings may be due to interrelated factors concerning genetic counseling, environmental factors, and increased anticipatory anxiety. Moreover, pregnant women have requested
additional information regarding the detailed prognosis of individuals with Down syndrome or other congenital disorders and/or artificial abortion, assuming positive NIPT results. Therefore, genetic counseling and appropriate follow-up sessions are essential for the well-being of pregnant women taking the NIPT.

Practice implications

To reduce internal conflict in pregnant women due to ambivalent feelings, it is critical to actively provide them detailed information about living with a child affected by one of the disorders screened during the genetic counseling sessions and to discuss this information assuming a potentially positive result, including specifics regarding medical pregnancy termination. Importantly, we believe that pregnant women should receive personalized psychological care so that they can make independent decisions based on factual knowledge. Thus, even if pregnant women experience feelings of ambivalence, they should be reassured and accept that undergoing NIPT was their decision after careful consideration, thereby reducing their potential internal conflicts.

Appendix

Table 1

Table 2

Abbreviation

NIPT: Non-invasive prenatal testing

Declarations
Acknowledgement

We thank all clinical geneticists and genetic counselors who participated in this study for making this study possible. We thank all the respondents participating in this study.

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Author Contributions

J. Y.: Study planning,
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All authors contributed to and have approved the final manuscript.

Funding

Junko Yotsumoto was supported by the Grant-in-Aid for Scientific Research (C), Japan

The funding source had no role in designing or conducting the study, nor in the collection, management, analysis or interpretation of data; in the preparation, review or approval of the manuscript; or in the decision to submit the paper for publication.

Ethics approval and consent to participate

Subjects have given their written informed consent.

The study protocol has been approved by the ethics committee of Center for Maternal-Fetal, Neonatal and Reproductive Medicine, National Center for Child Health and Development(No 602), and all research institute’s committee on human research.

Consent for publication

Not applicable.
Competing interests

The authors have no conflicts of interest to declare.

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Tables

Due to technical limitations, tables 1 and 2 are only available as a download in the supplemental files section.

Figures
Fig. 1. Flowchart of the respondent selection

Flowchart of the respondent selection
Figure 2

Factors associated with ambivalence Footnote: n = Number of occurrences of each category. Categories may partially overlap.

Supplementary Files

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