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Women's and health professionals' preferences for prenatal tests for Down syndrome: a discrete choice experiment to contrast noninvasive prenatal diagnosis with current invasive tests

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Purpose: To compare the preferences of women and health professionals for key attributes of noninvasive prenatal diagnosis for Down syndrome relative to current invasive tests.

Methods: A questionnaire incorporating a discrete choice experiment was used to obtain participants' stated preference for diagnostic tests that varied according to four attributes: accuracy, time of test, risk of miscarriage, and provision of information about Down syndrome only or Down syndrome and other conditions. Women and health professionals were recruited from five maternity services in England and a patient support group.

Results: Questionnaires from 335 women and 181 health professionals were analyzed. Safe tests, conducted early in pregnancy, with high accuracy and information about Down syndrome and other conditions were preferred. The key attribute affecting women's preferences for testing was no risk of miscarriage, whereas for health professionals it was accuracy.

Conclusions: Policies for implementing noninvasive prenatal diagnosis must consider the differences between women's and health professionals' preferences to ensure the needs of all stakeholders are met. Women's strong preference for tests with no risk of miscarriage demonstrates that consideration for safety of the fetus is paramount in decision making. Effective pretest counseling is therefore essential to ensure women understand the possible implications of results.

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Key Words: discrete choice experiment; Down syndrome; non-invasive prenatal diagnosis

INTRODUCTION

Currently in the United Kingdom, the National Screening Committee recommends that all pregnant women be offered a screening test for Down syndrome. Women who have a "high-risk" result are then offered an invasive diagnostic test, chorionic villus sampling or amniocentesis, which will tell them definitively whether or not the baby has Down syndrome. These invasive tests carry a risk of miscarriage of around 1%[1] and cannot be performed until 11 weeks of pregnancy. The possibility of an alternative, noninvasive approach based on a maternal blood sample was raised with the discovery of cell-free fetal DNA in the maternal circulation more than a decade ago.[2] Cell-free fetal DNA can be detected from 4 to 5 weeks gestation[3] and is rapidly cleared from the maternal circulation after delivery, making it pregnancy specific.[4] The main barrier to developing prenatal tests based on cell-free fetal DNA has been in distinguishing the information specific to the fetus, as the majority of cell-free DNA in the circulation is maternal in origin.[5] The technology underpinning noninvasive prenatal diagnosis (NIPD) for Down syndrome has advanced rapidly and several large scale validity studies have now been conducted to evaluate testing based on massively parallel sequencing.[6–8] Moreover, the first NIPD tests for Down syndrome are now available commercially in some countries[9] and considerations for more widespread implementation are being debated.[10] Validation of NIPD for Down syndrome using sequencing approaches is, however, ongoing and the test is not yet considered fully diagnostic as the small, but significant rate of false positives means an invasive test is still required to confirm a positive result.[11]

The clinical benefit of a NIPD test using cell-free fetal DNA is clear, as the risk of miscarriage associated with invasive testing is avoided. However, decisions on when and how NIPD for Down syndrome is introduced need to include considerations regarding other attributes of the test such as its accuracy and timing in pregnancy as it may be possible to perform NIPD several weeks earlier than current invasive tests. Another key difference between NIPD and invasive tests lies in the amount of information available from test results, with NIPD likely to provide targeted information (trisomy 21 only or trisomies 21, 18, and 13 only) as compared with an invasive test where information on all chromosomes can be provided through karyotyping.

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Understanding which test attributes are most highly valued by consumers and health professionals will help guide the implementation of NIPD into National Health Service (NHS) practice in a way that best meets the needs of all stakeholders.

Discrete choice experiments (DCEs) have been widely used in health-care research to examine stakeholder preferences.[12] In a standard DCE, a series of hypothetical health-care options are presented and participants are asked to choose between them. As a result, DCEs reflect the complex nature of real-life decisions, allowing an exploration of people's preferences and their willingness to trade off one attribute against another.[13] DCEs have been used previously to examine preferences for screening and diagnostic tests for Down syndrome, looking at attributes such as miscarriage risk, the timing of the test, and the type of information available from test results.[14–18] Of note, in these studies a range of miscarriage risks were presented, but the option of a test with no risk of miscarriage was not included, which makes them inappropriate to assess preferences for NIPD. The results of the DCEs have had important implications for service delivery. For example, Bishop et al.[14] found that health professionals placed greater value on earlier tests in pregnancy than women who would in fact prefer to wait for a result until later in pregnancy if the test were safer and more accurate. The aim of this study was to compare the preferences of women and health professionals for key attributes of diagnostic tests for Down syndrome, taking into account a range of possible clinical features of NIPD relative to invasive tests.

MATERIALS AND METHODS

Ethics approval was obtained from the National Research Ethics Service Committee (London-Hampstead REC Reference: 11/H07201/12). Study design and analysis followed current guidelines for conducting DCEs.[13,19,20]

Study sample

Pregnant women were recruited from the obstetric ultrasound department London Hospital and three at regional NHS hospitals. This group was a convenience sample of English-speaking women who were attending for a second- or third-trimester ultrasound scan. All of the pregnant women invited to participate had either declined Down syndrome screening or had already undergone screening and had been given a low-risk result. Pregnant women with a high-risk result were not included in this study to avoid any interference with their personal decision making regarding prenatal diagnosis. Women were given the questionnaire and had the option of reading two brief information sheets (see **Supplementary Materials** and **Methods** online). The first sheet described Down syndrome, current tests for Down syndrome screening and diagnosis, and future noninvasive tests; the second sheet described test accuracy. A researcher was available to discuss any questions. Women were asked to complete the questionnaire while waiting for their scan appointment. If there was insufficient time for this, they were invited to complete the questionnaire at home and return it by reply-paid post.

To broaden the range of opinions, pregnant and nonpregnant women who were members of the patient support group Antenatal Results and Choices (ARC) were invited to participate. The ARC membership includes women who have had to make decisions about invasive testing following a high-risk result as well as having to decide on management options following a diagnosis of Down syndrome. Women were recruited via an advertisement on the ARC website, which asked them to contact the researchers if they were interested in the study. The questionnaire was then sent by post with a prepaid envelope for returning it. Questionnaires were also available at the ARC Annual General Meeting in September 2011 where interested members were invited to take a questionnaire to complete and return by prepaid post.

Health professionals, primarily midwives and obstetricians, delivering antenatal care to women and who were likely to discuss options for Down syndrome screening and diagnostic testing were recruited from one London and four regional NHS hospitals. At all the hospitals, potential participants were approached in person and invited to complete a hard copy of the questionnaire. At one hospital, relevant staff groups were also e-mailed a study invitation and had the option of completing an online questionnaire.

Questionnaire design

Attributes for the DCE component of the questionnaire were selected after a literature review of prenatal testing for Down syndrome, which included several previously published DCEs.[14–18] The chosen attributes into account key differences between NIPD and invasive tests and the associated levels clinically feasible ranges (**Figure 1a**). The DCE design follows the approach of Street and Burgess.[21] Two attributes had three levels and two attributes had two levels. The number of possible combinations of attributes and levels was statistically reduced from 32 ($2^3 \times 2^2$) to 9 scenarios using an orthogonal fractional main effects design[22] to give a practical number of choices for participants to complete in the questionnaire. A shift of one level was applied to the initial nine scenarios to create nine additional scenarios that were randomly paired to form the choice sets (see **Supplementary Materials** and **Methods** online). Across the choice sets, all levels of each attribute with equal frequency (level balance) and within each individual choice set there no overlap in attribute levels (minimal overlap). The statistical design was efficient in terms of maximizing the amount of information from respondents, given the number of attributes and levels (D-efficiency = 99.79%).[23] An additional question was included as an internal consistency check, with a clearly superior test as an option, giving a total of 10 pairwise choice sets (see **Supplementary Materials** and **Methods** online). The questionnaires for women asked about which test they would prefer to have and the questionnaires for health professionals asked about which test they would prefer to offer. Participants were asked to choose test A, test B, or neither (**Figure 1b**). The neither option was included to make the

a

Attribute	Levels
Accuracy	90%, 95%, 100%
Time of results (gestation in weeks)	9, 11, 13
Risk of miscarriage	Small risk (1%), No risk
Information gained from the test	Down syndrome, Down syndrome plus additional information

b

Choice 1	Test A	Test B
Accuracy	95%	100%
Time of results	9 weeks	11 weeks
Risk of miscarriage	Small risk (1%)	No risk
Information from test	Down's syndrome plus additional information	Down's syndrome only

Which test would you prefer (*tick one box only***)?**
Test A ☐ Test B ☐ Neither ☐

Figure 1 Discrete choice experiment design. (**a**) Attributes and levels used in the discrete choice experiment. (**b**) Example of a discrete choice experiment choice set.

choice more realistic, as in practice women may choose not to have a test.

The questionnaires included a description of the attributes and levels included in the DCE and a section asking participants to rank the four attributes in order of importance. Additional questions for women included age, ethnicity, education, parity, gestational age, and uptake of Down syndrome screening. Additional questions for health professionals included job title, years in role, age, and gender. The questionnaires took ~20 min to complete. Questionnaires were piloted with 17 midwives and 20 women to determine whether they could be readily understood; participants were asked if there were any other important attributes of prenatal tests that were not covered in the questionnaire. Following this pilot, changes were made to the wording in the questionnaire describing the attributes, but not to the overall design of the experiment.

Analysis

The DCE preference data were analyzed for both women and health professionals using a conditional logit regression model.[24] For data entry, to avoid misinterpretation[19] the levels for accuracy and time of results coding were mean centered, and risk of miscarriage and information were effects coded. A constant term was included to reflect the "neither" option.[25] The sign (+ or −) of the coefficients generated in the regression analysis indicates the direction of the preference for each attribute. We anticipated positive coefficients for three attributes as we expected participants to prefer tests with greater accuracy, no risk of miscarriage, and comprehensive information. We anticipated a negative coefficient for the timing attribute, which would suggest preference for an earlier test.

The preferences of women were compared with those of health professionals. Additional subgroup analyses were performed to examine differences related to women's age (<35 or ≥35), screening uptake, how they were recruited (via ARC or in maternity units); and for health professionals, differences between midwives and obstetricians. In addition, we determined those women and health professionals who considered multiple attributes when choosing between tests ("traders") and those who made choices on the basis of one attribute only ("nontraders"). We then compared the women and health professionals who were "traders."

To explore the trade-offs the participants were willing to make between test attributes, we calculated the marginal rates of substitution. The marginal rates of substitution between two attributes is the ratio of their coefficients. This allows a direct assessment of how much of one attribute the participants are willing to trade for one unit of another attribute, and permits an easy comparison between different attributes on a common scale.[19] We also used the regression results to calculate the predicted probability that tests with different combinations of the attributes and levels used in the choice sets would be selected. This allowed us to rank the tests in terms of their order of preference by the participants.[19]

The software package Stata 10.0 (StataCorp College Station, TX) was used to perform all analyses.

RESULTS

Participants

The response rate was 94.9% (318/335) for women recruited through maternity units and 61.5% (32/52) for those recruited through ARC. The response rate for health professionals was 54.5% (193/354). Questionnaires were excluded if the consistency question was not answered as expected (women $n = 5$, and health professionals $n = 8$) or if the respondents did not complete the choice set (women $n = 10$, and health professionals $n = 4$). Consequently, questionnaires from a total of 335 women and 181 health professionals were included in the analysis. Demographic information is summarized in **Table 1**.

Table 1 Demographic data

Women	**Total (*n* = 335)[a]**
Recruiting center	
Hospital 1	78 (23.28%)
Hospital 2	37 (11.04%)
Hospital 3	98 (29.25%)
Hospital 4	90 (26.87%)
ARC	32 (9.49%)
Age	
Mean (SD)	30.94 (6.29)
Ethnicity	
White or white British	297 (88.6%)
Black or black British	8 (2.39%)
Asian, East Asian, or Asian British	15 (4.48%)
Other	10 (2.99%)
Highest qualification	
No qualification	8 (2.39%)
GCSE or equivalent	71 (21.19%)
A-level or equivalent	63 (18.81%)
Degree or equivalent	184 (54.93%)
Number of children	
None	163 (48.66%)
1	112 (33.43%)
2 or more	58 (17.31%)
Do you have a child with Down syndrome?	
Yes	0
No	327 (97.6%)
Does anyone you know have a child with Down syndrome?	
Yes	75 (22.39%)
No	252 (75.22%)
Current pregnancy	
Currently pregnant	302 (90.15%)
Gestation (weeks)—mean (SD)	24.86 (7.07)
Down syndrome screening in this pregnancy	
Have had or will have screening	245 (81.13%)
Do not intend to have screening	55 (18.21%)
Health professionals	**Total (*n* = 181)[a]**
Recruiting center	
Hospital 1	26 (14.36%)
Hospital 2	19 (10.50%)
Hospital 3	42 (23.20%)
Hospital 4	33 (18.23%)
Hospital 5	61 (33.70%)
Age	
Mean (SD)	40.12 (9.16)
Gender	
Female	153 (84.53%)
Male	25 (13.81%)
Profession	
Obstetrician	53 (29.28%)
Midwife	117 (64.64%)
Other (sonographer, nurse consultant, etc.)	9 (4.97%)
Years in profession	
<5	48 (26.51%)
6–15	58 (32.04%)
16–25	50 (27.62%)
26–35	17 (9.39%)

ARC, Antenatal Results and Choices; GCSE, General Certificate of Secondary Education.
[a]Values reported as *n* (%) unless otherwise stated. Totals may not add to 100% due to missing responses.

Ranking of attributes

Ranking of attributes varied between participants with 66% of women ranking safety highest, whereas 58% of health professionals ranked accuracy as the most important (see **Supplementary Results** online).

Regression results

The positive coefficients suggest that both women and health professionals prefer a test with greater accuracy, no risk of miscarriage, and full information (Table 2A). The negative coefficient for time of results indicates the preference for an earlier test. These results support the *a priori* expectations providing support for the theoretical validity of the models. All coefficients were statistically significant for both groups. Comparison of regression results for professionals and women confirmed the differences observed from rankings with statistically significant differences in the coefficients for accuracy, timing, and no risk of miscarriage (Table 2A).

Further subgroup comparisons indicated that women aged ≥35 years placed a greater emphasis on accuracy than younger women, and women who had undergone Down syndrome screening in their current pregnancy differed from those who had declined Down syndrome screening in the emphasis they placed on test timing and information (Table 2B). Women recruited through ARC were more likely to choose tests with greater accuracy and full information (Table 2B). There were no significant differences in preferences between obstetricians and midwives (see **Supplementary Results** online).

Of the respondents defined as nontraders, 16 (4.78%) women, but no health professionals chose neither for all options; 110 (32.84%) women and 13 (7.18%) health professionals chose tests based on no miscarriage risk; 15 (4.48%) women and 33 (18.2%) health professionals chose tests based on highest accuracy; 1 (0.30%) woman and no health professionals chose tests based on earliest timing; and 19 (5.67%) women and 7 (3.87%) health professionals chose tests based on full information being available. When comparing traders, the only significant difference between the groups was for accuracy (Table 2B).

Table 2 Conditional logit analysis regression results

A. For women and health professionals

	Women (n = 335)		Health professionals (n = 181)		Difference
Attribute	**Coefficient (95% CI)[a]**	***P* value**	**Coefficient (95% CI)[b]**	***P* value**	***P* value**
Accuracy	0.101 (0.087 to 0.116)	<0.0001	0.265 (0.240 to 0.289)	<0.0001	<0.0001
Time of results	–0.086 (–0.117 to –0.055)	<0.0001	–0.144 (-0.188 to –0.100)	<0.0001	0.0375
Full information	0.746 (0.642 to 0.850)	<0.0001	0.834 (0.691 to 0.977)	<0.0001	0.3170
No miscarriage risk	1.627 (1.516 to 1.738)	<0.0001	1.061 (0.900 to 1.223)	<0.0001	<0.0001

B. For subgroup comparisons

	Women's age			Women's uptake of Down syndrome screening			Women's recruitment location			Participants willing to trade		
	Coefficient			Coefficient			Coefficient			Coefficient		
Attribute	**<35[c] (n = 234)**	**≥35[d] (n = 97)**	**Difference (*P* value)**	**Yes[e] (n = 245)**	**No[f] (n = 55)**	**Difference (*P* value)**	**ARC[g] (n = 32)**	**Maternity care[h] (n = 303)**	**Difference (*P* value)**	**Women[i] (n = 174)**	**HP[j] (n = 128)**	**Difference (*P* value)**
Accuracy	0.092	0.127	0.0395	0.101	0.090	0.5731	0.162	0.096	0.0145	0.131	0.259	<0.0001
Time of results	–0.086	–0.089	0.9498	–0.100	–0.014[k]	0.0492	–0.121[l]	–0.083	0.4954	–0.136	–0.171	0.3173
Full information	0.755	0.721	0.7601	0.755	0.442	0.0270	1.358	0.690	0.0012	0.855	0.872	0.8758
No miscarriage risk	1.674	1.510	0.1732	1.651	1.709	0.7141	1.395	1.657	0.2185	1.123	1.085	0.2644

ARC, Antenatal Results and Choices; CI, confidence interval; HP, health professionals.
[a]Number of observations = 9,975; pseudo-R^2 = 0.3557; [b]Number of observations = 5,427; pseudo-R^2 = 0.4201; [c]Number of observations = 7,020; pseudo-R^2 = 0.3467; [d]Number of observations = 2,838; pseudo-R^2 = 0.3765; [e]Number of observations = 7,407; pseudo-R^2 = 0.4230; [f]Number of observations = 1,620; pseudo-R^2 = 0.1985; [g]Number of observations = 921; pseudo-R^2 =0.3520; [h]Number of observations = 9,054; pseudo-R^2 = 0.3610; [i]Number of observations = 5,144; pseudo-R^2 = 0.3246; [j]Number of observations = 3,837; pseudo-R^2 = 0.3899; [k]Coefficient not significant; [l]Coefficient significant at P <0.05; all other coefficients significant at P <0.0001.

Marginal rates of substitution

Calculation of the marginal rates of substitution confirmed women's strong preference for a test with no risk of miscarriage, as they were prepared to wait more than twice as long and accept 12% lower accuracy for a test that had no risk of miscarriage as compared with health professionals (**Table 3**).

Predicted probabilities

Tests were ranked in order of preference by calculating the mean probability of choosing a given test (**Table 4A**). Two tests with attributes similar to current invasive tests were included in the analysis for comparison with a series of tests with the possible attributes of NIPD. The rankings show that women are prepared to accept tests with lower accuracy if there is no risk of miscarriage (**Table 4A**), whereas health professionals prefer to offer a test that is accurate, even if it has a small risk of miscarriage (**Table 4B**).

DISCUSSION

The introduction of NIPD for Down syndrome into routine clinical practice is imminent. Implementation of any test needs to consider more than laboratory robustness and should take account of the needs and preferences of service users and providers in order to ensure the development of appropriate care pathways. In this study, we have examined the preferences of women and health professionals for diagnostic tests for Down syndrome. Not surprisingly, both women and health professionals were found to prefer safe tests that were conducted early in pregnancy with high accuracy and gave information about other conditions in addition to Down syndrome. However, there were significant differences between the women and health professionals in the relative values they placed on safety, accuracy, and time of results, with women prepared to wait longer and accept lower accuracy if the test had no risk of miscarriage.

Table 3 Marginal rates of substitution

	Number of weeks respondents are prepared to wait		Reduction in accuracy (%) respondents are prepared to accept	
	Women	Health professionals	Women	Health professionals
Test with no risk of miscarriage	18.92 (1.627/–0.086)	7.37 (1.061/–0.144)	16.11 (1.627/0.101)	4.00 (1.061/0.265)
Test with full information	8.67 (0.746/–0.086)	5.79 (0.834/–0.144)	7.39 (0.746/0.101)	3.15 (0.834/0.265)
Test with 5% greater accuracy	5.87 (0.101/–0.086 ×5)	9.20 (0.265/–0.144 ×5)	—	—

Table 4 Probability analysis

Ranking	Mean probability	Option	Accuracy (%)	Time (weeks)	Information	Risk of miscarriage
A. For women						
1	0.9281	Perfect test	100	9	Full	No risk
2	0.8067	NIPD option 2	100	9	DS only	No risk
3	0.6992	NIPD option 1	100	11	DS only	Small risk
4	0.5550	NIPD option 4	95	11	DS only	No risk
5	0.5290	NIPD option 5	90	9	DS only	No risk
6	0.4480	NIPD option 3	95	13	DS only	No risk
7	0.4340	IPD (early)	100	9	Full	Small risk
8	0.3218	IPD (late)	100	13	Full	Small risk
9	0.0242	Worst test	90	13	DS only	Small risk
B. For health professionals						
1	0.9729	Perfect test	100	9	Full	No risk
2	0.9152	NIPD option 2	100	9	DS only	No risk
3	0.7993	IPD (early)	100	9	DS only	Small risk
4	0.7406	NIPD option 1	100	11	DS only	No risk
5	0.6417	IPD (late)	100	13	Full	Small risk
6	0.2862	NIPD option 4	95	11	DS only	No risk
7	0.2531	NIPD option 5	90	9	DS only	No risk
8	0.1502	NIPD option 3	95	13	DS only	No risk
9	0.0068	Worst test	90	13	DS only	Small risk

DS, Down syndrome; IPD, invasive prenatal diagnosis; NIPD, noninvasive prenatal diagnosis.

Previous DCE studies looking at prenatal testing for Down syndrome have also found differences between women's and health professionals' preferences. Studies conducted in the United Kingdom[14] and Australia[15] have shown health professionals valued earlier timing of tests whereas women emphasized safety. One possibility to account for the differences is that women may be less likely to look objectively at multiple test attributes and may not have the same understanding of the implications of the differences in attribute levels, as they are unlikely to think about tests involving their unborn baby in this way.[14,15] Health professionals are also more likely to be aware of the growing body of literature, exploring the ethical and psychosocial concerns associated with NIPD.[26–30] Consequently, the value placed on accuracy by health professionals as compared with women in this study may, at least in part, be due to differences in their existing knowledge and concerns about the implementation of NIPD. To an extent, this view is supported by the observation that, in contrast to women recruited through antenatal clinics, those recruited through ARC, all of whom had experience of adverse results in a previous pregnancy and hence prior experience, had a greater preference for accuracy and a test that gave more information. Experiential knowledge has previously been shown to play an important role in women's decisions regarding prenatal testing[31] and personal experience may account for the differences seen between these groups. We also found that women ≥35 had a stronger preference for tests with higher accuracy than women <35. This is similar to the observation of Mulvey et al.,[32] who found that older women (>37) valued Down syndrome screening tests with the highest detection rate, as compared to younger women, who preferred tests with the lowest possible false-positive rate.

Having a test with no risk of miscarriage was clearly a major consideration for women when making decisions about the prenatal tests with which they were presented. Of note, over one-third of the women in this study chose tests based only on the fact there was no risk of miscarriage and did not trade on the other test attributes. This finding corresponds with recent research from the United States that reported that pregnant women thought the most important feature of NIPD would be safety of the fetus (75%), followed by accuracy (13%) and early availability of results (7%).[33] Similarly, a UK study found that women who have had NIPD for fetal sex determination described safety as the most important feature of the test.[34]

For many women, the miscarriage risk associated with invasive testing is a psychological barrier to diagnostic testing for Down syndrome.[27,35] If the miscarriage risk is removed, women may feel more inclined to have a test.[35] In addition, health professionals have been found to view NIPD more like screening than an invasive diagnostic test, suggesting that they may not provide the same level of information and counseling as they would for an invasive diagnostic test.[36] The results of our study demonstrate that the option of a test with no miscarriage risk is indeed highly influential for women making decisions regarding prenatal testing. The predicted probability analysis shows that women ranked the attributes of the current invasive tests lower than the theoretical NIPD tests, regardless of test accuracy. In this study there were 50 pregnant women who had chosen not to have Down syndrome screening in their current pregnancy; of these only 12 indicated that they would not have any of the tests presented to them on the questionnaire by selecting "neither" for all of the choice options. This suggests that it is possible that many of the women who currently decline Down syndrome screening may choose to undergo testing if there is no risk of miscarriage associated with the definitive diagnosis. However, more research is needed to determine whether, if faced with a real-life decision, the uptake of Down syndrome testing would increase if NIPD was available.

Differences in the relative values women and health professionals place on test attributes are important considerations for the implementation of NIPD as health professionals play key roles in evaluating health innovations and establishing policy.[37,38] Ideally, the implementation of NIPD will take all stakeholder views into consideration. Furthermore, health professionals must recognize that their views about prenatal tests may differ from those of the women with whom they discuss testing. The attitudes of health professionals have been shown to impact the uptake of Down syndrome screening,[39] and in the recent study of pregnant women's interest in NIPD conducted in the United States, one in five women said that they would do what their health professional recommended.[33]

Implementation of NIPD for Down syndrome into routine antenatal care will depend on many factors, including test accuracy, costs, and care pathways.[10] A key factor for the successful introduction of NIPD will be the development of approaches to counseling and strategies for information provision that will facilitate informed decision making. As safety is so clearly predominant in women's minds, care must be taken in counseling to spell out all the differences between available testing options or women may not think beyond the issue of safety and accept testing without realizing the possible implications of the results. For this reason, it will also be critical to have structures in place for when abnormal results are given to help parents to understand the clinical implications of the findings and to provide individualized support for decision making.[40,41]

Limitations

In this study we attempted to gather the preferences of a diverse cross-section of women by recruiting from four different hospitals in disparate regions of the United Kingdom. However, a number of issues may limit the generalizability of our findings. For example, the overwhelming majority of pregnant women who took part in this study were white, and more than half were highly educated and held a degree qualification or equivalent. We considered only four attributes of prenatal tests. Real-life choices about prenatal tests also consider other factors such as false positives, access to tests, costs, and counseling. Consideration could also be given to whether preferences for diagnostic tests were influenced by having to choose between different types of procedures, for example,

a blood draw as compared with an invasive test, which is often associated with pain and anxiety.[42] Furthermore, cost is frequently included as an attribute in DCEs to estimate willingness to pay. Although cost will be an important consideration for future research in this area, at present the actual cost of NIPD is uncertain. The technology underpinning NIPD for Down syndrome is rapidly becoming cheaper. In addition, a key determinant of the cost of NIPD will be how it is placed in the care pathway, as this will establish how many people are offered testing.[10] Furthermore, in this study we were considering implementation into routine NHS care, a service that does not charge for testing.

It is also important to note that, as for any stated preference study, the choices made in the questionnaire do not necessarily reflect the choices that would be made if participants were faced with a real-life decision about testing. Most of the pregnant women completing the questionnaire were at 20 or more weeks gestation and were invited to participate as they had either chosen not to have Down syndrome screening or were found to be at low risk. As such, their stated preferences may be different from the choices they would make earlier in pregnancy or if they had received a high-risk screening result. Finally, the DCE design does not look into the reasoning behind the choices made or give insight into how the tests were perceived. An in-depth understanding of the thoughts of pregnant women and preferences regarding NIPD is needed and this could be obtained through qualitative studies.

Conclusions

Women and health professionals differ in the value they place on test accuracy and safety when making choices about diagnostic tests for Down syndrome. Policies for the implementation of NIPD for Down syndrome need to consider these differences to ensure the needs of all stakeholders are met. Women's strong preference for tests with no risk of miscarriage demonstrate that consideration for the safety of the fetus has the potential to outweigh other test attributes when making decisions regarding prenatal testing. This highlights the need for effective pretest counseling and informed consent processes to ensure women understand the parameters of the test and the possible implications of NIPD.

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DISCLOSURE

The authors declare no conflict of interest.

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