ORIGINAL ARTICLE

Antenatal thalassaemia carrier testing: women's perceptions of 'information' and 'consent'

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Objectives: To explore the attitudes of a sample of pregnant women in the UK towards informed consent for antenatal thalassaemia carrier testing and perceived pre-test information needs for such testing.

Setting: The study was conducted in two cities in the North of England, where participants were recruited via Midwifery and Genetic services.

Method: In all, 110 Pakistani women tested and not found to be thalassaemia carriers completed a questionnaire, 14 of whom were also interviewed. Thirty-six women identified as carriers or possible carriers completed a questionnaire and were interviewed. The questionnaires assessed whether women were aware that they had been tested for thalassaemia carrier status, whether they were asked for their consent for such testing, and their pre-test information preferences. The interviews explored women's beliefs about 'informed consent' in more depth.

Results: Women had received little or no pre-test information and said that they would have preferred to be informed that they were being tested, but they did not expect, or express a desire, to be asked for their informed consent.

Conclusion: While information was important to women, consenting was not. Overall, women discussed 'information' and 'consent' as two separate issues, thus challenging assumptions around the term informed consent. Women wanted pre-test information because they wanted to know more about the tests that they would be having, not to use it to make decisions about whether to have the tests.

INTRODUCTION

Following recognition of the potential adverse psychological consequences after receipt of test results and changing social attitudes, the National Screening Committee believes that screening programmes should offer the choice of screening tests to individuals.[1] Guidelines recommend that 'adequately informed consent should be a requirement for all genetic screening programmes'.[2,3]

There is no universal definition of 'informed consent', although there is general agreement in the literature that it has two core characteristics: firstly, that the decision to accept or decline the test is based on good-quality information; and secondly, that it is consistent with the decision-maker's values.[4,5] There is also little agreement on the amount of pre-test information that should be provided. Suggestions for pre-test information include the following:[2,6–8]

- the purpose of the screening test;
- a description of the genetic condition, including its severity, variability and therapeutic options;
- the genetic inheritance pattern of the condition;
- the reliability of the screening test, including false positives and false negatives;
- the procedure for informing individuals of both negative and positive results;
- the implications of a positive result for their future and existing children, and for other family members;
- an explanation of the subsequent options, such as testing partner, prenatal diagnosis (chorionic villus sampling or amniocentesis – both have 1–2% risk of miscarriage), termination/continuation with pregnancy;
- a warning for pregnant women that genetic screening tests may reveal information about paternity;
- the uncertainties and risks attached to the screening process;
- any significant medical, social or financial implications of screening tests.

The literature also suggests that this information should be presented in a neutral, non-directive manner, allowing individuals to assimilate the information in accordance with their own values.[6,7] In addition, 'adequately informed consent should be a requirement for all genetic screening programmes and to help people decide whether or not to be screened, it is important to provide both written and oral information in a language appropriate to the individual'.[2]

Thalassaemia

β-thalassaemia major (thalassaemia) is a serious recessive genetic condition that is prevalent among people of Mediterranean, Middle Eastern and South Asian (Indian, Pakistani, Bangladeshi) origin. Individuals with thalassaemia are unable to make sufficient and sustainable adult haemoglobin (Hb), so they require regular monthly blood transfusions. This unfortunately results in excess iron, which has to be removed daily by injecting a drug called Desferal slowly under the skin for 8–12 h using a pump-driven syringe.[9,10] Generally, thalassaemia is not a curable condition, although bone marrow transplantation offers the possibility of cure to those who have an immunologically matched sibling.[11] The birth of a child with thalassaemia can be prevented if 'at-risk' couples opt for prenatal diagnosis

and subsequently opt for termination of an affected fetus. At-risk couples are those where both parents are carriers of the faulty gene, and, on average, one in four of their pregnancies will be affected. Ideally, prospective parents from the Mediterranean, Middle East and South Asia would know before pregnancy whether or not they were carriers, but in practice this is generally not the case. Therefore, at the time of this study, selective antenatal thalassaemia carrier testing was being carried out in many parts of the UK, where local policies stated that midwives should routinely offer testing for Hb abnormalities to all pregnant women who are not of Northern European origin. If a woman was found to be a carrier, then her partner would also be offered carrier testing; she would not be at risk of having a child with thalassaemia unless both parents were carriers.

Thalassaemia carrier testing in practice

In order to understand how the various test results are derived and how possible issues about informed consent can arise, it is important to look at the laboratory diagnosis of thalassaemia carriers, which leads to the diagnosis of α-thalassaemia and β-thalassaemia carriers, and of other Hb variants. This process involves three stages (Figure 1).

Stage 1

This involves measuring the mean cell Hb level (MCH), where an MCH less than 27 per gram (pg) indicates the presence of a thalassaemia (α or β), iron deficiency, or both. If the MCH is less than 27 pg, further investigations are carried out to identify β-thalassaemia carriers (i.e. Stage 2). It is important to note that this first measure for identifying thalassaemia carriers forms part of routine antenatal blood investigations and further testing only takes place if this routine measure indicates an abnormality. This then means that all pregnant women are in fact screened for thalassaemia carrier status.[13] So, in selective antenatal testing, women of Northern European origin to whom testing has not been offered may nonetheless be tested for thalassaemia carrier status (Stages 2 and 3 in Figure 1), without having given prior consent, if they have an MCH less than 27 pg. Guidelines do not specify how to obtain consent for antenatal testing programmes where the initial indicator of an 'abnormality' is a routine measure.

Stage 2

Investigations at this stage can lead either to the diagnosis of β-thalassaemia carriers or to inconclusive results which require further investigations. Stage 2 involves measuring the HbA_2 level, where a value over 3.5% confirms β-thalassaemia carrier status. However, inconclusive results are produced at this stage if the HbA_2 level is less than or equal to 3.5%, indicating the presence of iron deficiency, α-thalassaemia, or rare forms of β-thalassaemia, which can only be diagnosed following DNA analysis. Given that individuals with inconclusive results could be thalassaemia carriers, they are referred to from here onwards as 'possible carriers'.

Stage 3

This stage involves further investigations, including DNA analysis, which can take four to six weeks, so the expedient option is to assume that 'possible carriers' are in fact thalassaemia carriers and employ the same process for subsequent actions as for β-thalassaemia (i.e. offer carrier testing to their partners – even though these possible carriers are likely to be α-plus thalassaemia carriers, which has little clinical significance, or simply iron deficient).

At the NHS Trusts where the study was conducted, the midwives were operating the agreed selective testing protocol and understood that only women who were not of Northern European origin should be offered or informed about thalassaemia carrier testing, and believed that only a request from them would trigger the laboratory to perform such testing.[14] They were unaware that the MCH measure, routinely performed on all pregnant women for other purposes, formed the first stage of such testing, and that an abnormal MCH measure would automatically lead to further laboratory investigations for thalassaemia carrier status.

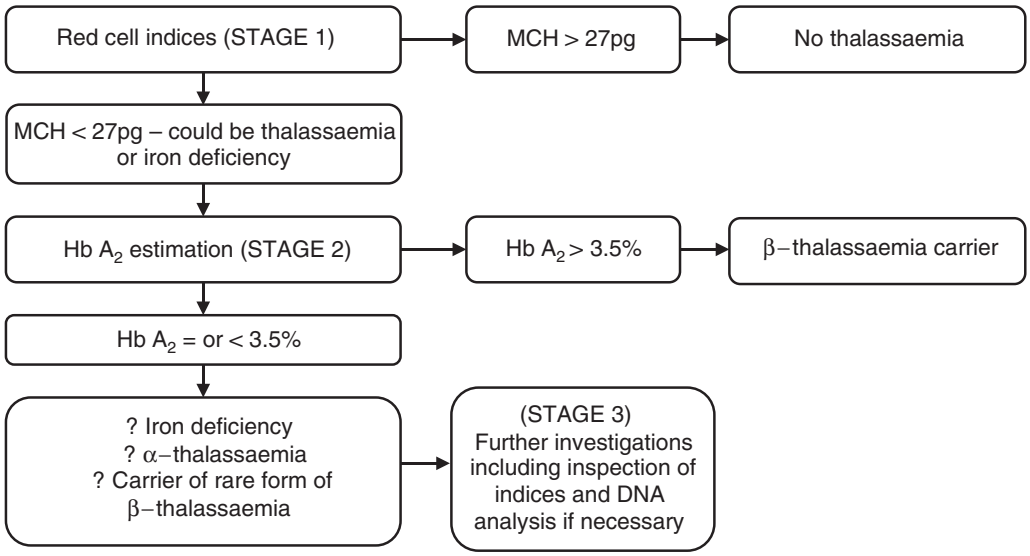

Figure 1 The laboratory diagnosis of thalassaemia carriers (adapted from Modell and Anionwu[12])

Assumptions associated with informed consent

There are a number of assumptions associated with the concept of informed consent. These include: people wish to be informed; people wish to make choices about their care; and people wish their consent to be sought. Literature on desire for information and desire to make decisions indicates that while people want more information on whatever medical procedure they are about to undergo, many people prefer health professionals to make decisions on their behalf.[15,16] Providing choices could result in women being faced with complex decisions. In addition, people are unlikely to become autonomous decision-makers and it could be considered unethical to force people to make decisions they may not want to make.[15] Nevertheless, bio-ethicists argue that people have a duty to be informed, to learn about themselves and their potential fate.[17] Rather than arguing about whether health professionals or patients should make decisions about genetic testing, others suggest that making such a decision in a medical context lies on a continuum and that the aim should be for mutual decision-making, or at least the patients' values being taken into account before making a decision on their behalf.[18] In addition, there is no evidence to show whether people from different communities share informed consent as a value. It may be that some people place greater emphasis on health professionals' judgements (i.e. 'They [health professionals] know best')[19] and prefer to trust health professionals to make health-related choices for them.[20]

Information is considered a prerequisite for antenatal testing for genetic conditions, but there is little guidance as to the content of pre-test information. Few studies have looked at people's views about informed consent; their perceived pre-test information needs for antenatal testing for genetic conditions; and whether and how people use pre-test information to consent to such testing. This paper describes the attitudes of a sample of pregnant women towards informed consent for antenatal thalassaemia carrier testing and their perceived pre-test information needs for such testing.

METHOD

Setting

The study was conducted in two cities in the North of England. Potential participants were accessed though Midwifery and Genetic services.

Design

The study included three groups of pregnant women who had been tested for thalassaemia carrier status and diagnosed as either β-thalassaemia carriers, possible thalassaemia carriers or not thalassaemia carriers. The original plan was to focus on Pakistani women because the hospitals involved in the study explained that they had a selective antenatal testing policy. However, on discovering that testing was universal in practice, a decision was taken to include all pregnant carriers and possible carriers of any ethnic origin.

Recruitment procedures

Approval was obtained from the relevant Local Research Ethics Committees. All carriers and possible carriers were recruited through thalassaemia counsellors, usually after they had received their partner's carrier test results. In City A, 26 women were identified as carriers over a period of 18 months (September 1999–March 2001). Twenty-two of these carriers were Pakistani, 15 of whom agreed to take part in the study. In City B, both pregnant Pakistani and white indigenous women were identified as thalassaemia carriers. Over a period of six months (1 August 2000–31 January 2001), 17 women were identified as β-thalassaemia carriers. Ten of these carriers were not approached to take part in the study either because they were not Pakistani or white indigenous women, or because their partner had not yet been tested (so had not been through the whole testing process). Four of the seven carriers approached agreed to take part in the study. Seventeen of the 36 possible carriers approached took part in the study: 15 of 23 asked on the telephone by the Antenatal Screening Coordinator and 2 of 13 who were asked by letter.

Over a period of eight months (February–September 2000), non-carriers were approached to take part in the study (initially by their midwife) after their second antenatal clinic visit, which is when they were provided with the results of their antenatal tests, including thalassaemia carrier status. At this stage, women should, in theory, have known which tests they had had and the results of these tests. Initially, the midwife briefly told the women about the study and introduced them to the researcher. Of the 125 non-carrier women who were given details about the study by the researcher, 110 agreed to take part in the study.

Data collection

All the women completed a questionnaire and all the carriers, possible carriers and 14 of the non-carriers were interviewed. The questionnaire asked pregnant women whether they were told and whether they were asked about thalassaemia carrier testing when their blood was taken for testing, and whether they would have wanted to know that they would be tested. Women were offered the choice of whether they wanted to complete the questionnaire in English or Urdu, and whether they wanted to complete the questionnaire themselves or with the aid of the researcher.

The researcher conducted all interviews in women's homes and in their chosen language (English, Urdu, Punjabi, Mirpuri or Hindu), using a guide developed from a review of the existing literature. Questions relating to informed consent explored:

- women's perceived pre-test information needs, including their reasons for their pre-test information preferences, the type, amount and timing of information preferred, and barriers to acquiring information;
- women's attitudes toward consent for antenatal thalassaemia carrier testing.

The interviews lasted between 30 and 60 min and were audiotaped with permission. Interviews were then translated where necessary and transcribed by the first author. Non-carriers completed the questionnaire either in the clinic immediately after their consultation with the midwife, or at home.

Analysis

The grounded theory approach was used for qualitative data collection and analysis, supported by N-Vivo (Nudist-Vivo 1.2; SAGE Publications). Interviews and data analysis were

conducted simultaneously to allow for exploration of emerging themes in subsequent interviews. Analysis showed that saturation was reached. The first author analysed the transcripts using the constant comparative method.[21] This involved coding data line by line to identify and assign codes to meaningful words, phrases or sentences. Codes were also given descriptions to allow comparison between them. Axial coding was carried out in parallel to open coding, which resulted in clustering codes with similar concepts into categories. The clustering of categories produced higher-order categories, which in turn were combined to develop themes to explain phenomena.

The findings for the three groups are presented together to provide an in-depth explanation of phenomena; that is, by including the perspective of pregnant women with different outcomes following antenatal thalassaemia carrier testing. The quantitative and qualitative findings are also presented together. Most of the findings in the Results section have been derived from the qualitative analysis. The quantitative results consist of percentages and frequencies and are presented at the beginning of subsections. All names used in the Results and Discussion sections are pseudonyms.

Table 1 Participants' demographic details

	Carriers ($n=19$)	Possible carriers ($n=17$)	Non-carriers ($n=110$)
Age (years)			
Median	24	30	23
Range	16–43	19–38	17–37
Gestation (weeks)			
Median	20	19	22
Range	9–33	12–32	17–34
Parity (number of women)			
Primiparous	12	4	85
Multiparous	7	13	25
Women's ethnic origin (number of women)			
Pakistani	19	10	110
Indigenous White	—	5	—
African-Caribbean	—	2	—
Women's self-ratings for quality of spoken English (number of women)			
Very fluent	7	9	47
Fluent	—	1	13
Okay	3	1	15
Poor	—	3	9
No English	9	3	26

RESULTS

Sample

The participants were 146 pregnant women tested for thalassaemia carrier status (see Table 1 for participants' demographic details). Nineteen of these had been diagnosed as carriers, 17 as possible carriers and 110 as non-carriers.

During the interviews, questions were asked separately about information and consent, which is reflected in the presentation of the results.

Provision of pre-test information for thalassaemia carrier testing

The researcher told women that the routine antenatal blood tests included testing for thalassaemia carrier status and asked them whether they had been told about such testing. Of the 146 women asked, 113 said they had not been told about thalassaemia carrier testing (77.4%) and 97 of these women (85.8%) said that they would have wanted to be told that they would be tested (Table 2).

The themes that emerged from the grounded theory analysis include perceived information needs, the relationship between information and anxiety, information preferences, barriers to obtaining information and attitudes towards consent for antenatal thalassaemia carrier testing.

Perceived pre-test information needs for antenatal thalassaemia carrier testing

Some women said that they wanted to be informed about conditions for which they had been tested so that they knew what their baby was not at risk of having.

> *Huma* (non-carrier, completed questionnaire in Urdu – 19 years old primigravida, six months in the UK): 'I was worried about why they took the blood. I should have been told what for. It's obvious that they're taking blood for seeing if there are any disorders, but we don't know what for.'

Women particularly wanted pre-test information if they had relatives with thalassaemia or were aware of their chances of being a thalassaemia carrier through the media,

Table 2 Whether women were told and whether they wanted to be told about thalassaemia carrier testing

	Groups			
	Carriers ($n=19$)	Possible carriers ($n=17$)	Non-carriers ($n=110$)	Total ($n=146$)
Told about thalassaemia carrier testing?				
Yes	1	4	22	27
No	13	13	87	113
Not applicable*	5	—	—	5
Do not know/cannot remember	—	—	1	1
Wanted to be told about thalassaemia carrier testing?				
Yes	12	9	76	97
No	2	4	11	17
Not applicable**	5	4	23	32

*Women who initiated the conversation about thalassaemia carrier testing, because they were aware of their carrier status.
**Women were not asked this question if they had said that they were told about thalassaemia carrier testing, or if they said that they did not know or could not remember whether they had been told

so that they could discount the possibility of their baby having thalassaemia.

A number of women also felt that they had an ethical right to know that they were being tested.

> *Jackie* (possible carrier – 26 years old, multigravida, White British): 'Ethically, is it right to test people for something that they're not aware that they're being tested for? … if it is being tested for, then surely women should have the right to know about it.'

Relationship between pre-test information and anxiety

Some women mentioned that provision of pre-test information could result in anxiety about conditions that the baby could have. A possible carrier said that she was not interested in pre-test information because 'you start thinking too much'. On the other hand, a number of women who also acknowledged that such information could increase anxiety said that they would have wanted pre-test information. For example, Fozia suggested that women worry about their baby's wellbeing regardless of what they are told about tests. So providing pre-test information does not make them worry more, it just gives them something else to think about. She also believed that such worry was not harmful.

> *Fozia* (non-carrier, interviewed in English – 20 years old, primigravida, born in the UK): '… I mean it's not as if we're not worried anyway. So something like that [pre-test information], I don't think it would have mattered to me even if I was worried about it because I do worry, worry all through my pregnancy.'

Many women suggested that pre-test information should be provided regardless of whether it causes anxiety. Instead of being perceived as harmful for the mother, anxiety was perceived as an inevitable, but natural, part of being pregnant. Another woman suggested:

> *Jackie* (possible carrier – 26 years old, multigravida, White British): '…you need to be told all the information … it does cause probably more distress …but that's just something you've got to go through. It's part of being pregnant.'

Pre-test information was also seen as a means of reducing anxiety on receipt of a positive test result. Most of the carriers and some of the possible carriers said that information about being tested would have forewarned them that they could receive a positive test result and may have prepared them so that they were not as 'shocked' by the result as they were when they were unaware of being tested:

> *Naheed* (β-thalassaemia carrier, interviewed in Mirpuri – 21 years old, primigravida, 10 months in the UK): 'If people know about the test and what a thalassaemia carrier is then they wouldn't be so worried when they find out that they are a carrier.'

Women's positive thalassaemia carrier results were accompanied by an unexpected urgent request for their husband to have a blood test. This may have increased women's perceptions of the severity of their positive test results:

> *Nageena* (β-thalassaemia carrier, interviewed in English – 24 years old, primigravida, born in the UK): 'you must immediately come to the hospital and get yourself tested and get your husband tested… And it's really frightening is that. I was upset and I was frightened.'

Women felt that pre-test information alerting them about the implications of a positive test result would have resulted in comparatively less anxiety than knowing nothing about the test or the possibility of receiving such results.

Type, amount and timing of information preferred

On the questionnaire for non-carriers, the majority of women indicated that they wanted pre-test information to include details about the condition (81%), the test procedure (80%), when the results would be available (80%), the meaning of positive and negative results (73%), and likely action following a positive result (77%). However, during the interviews many women suggested that they did not want pre-test information to include 'too much detail'. The different groups of women interviewed showed a preference for different amounts and type of information at different stages in pregnancy.

During the interviews, the non-carriers were less likely to say that they wanted detailed pre-test information. For example, some of them did not want any information, saying that they were happy knowing that 'everything was fine', while some said that pre-test information should include details about the condition for which they were being tested, why they were being tested and the possible outcomes of testing.

Carriers and possible carriers wanted more detailed pre-test information than the non-carriers. They suggested that pre-test information should include details about receiving positive results via a thalassaemia counsellor or letter, and the implications of being a thalassaemia carrier. Many of these women clarified that they did not want too much pre-test information, because there was already a lot of information being exchanged during the booking session:

> *Fiona* (possible carrier – 32 years old, primigravida, White British): '…the midwife is getting a lot of information from you and vice versa so you don't want to be bombarded with too much information…'

Some of the women did not expect midwives to go into great detail for any specific antenatal test because of time constraints on the midwife:

> *Jackie* (possible carrier – 26 years old, multigravida, White British): 'I'm not suggesting that they have a discussion with every single woman who goes through antenatal testing. Obviously that takes up a lot of the time of the midwives, and obviously their time is precious, they have a lot of women in their care that they have to speak to.'

Some of the possible carriers highlighted that the issue of thalassaemia is not salient to them at the time of testing, so they are unlikely to be interested in detailed pre-test information. They also suggested that pre-test information could be best provided in the form of a leaflet which they could read immediately or refer to on receipt of a positive result. Women from all three groups suggested that the most appropriate time for detailed information and explanations was on the receipt of a positive test result. They suggested that information provided at this stage should include details about the condition, why the woman is a thalassaemia carrier, implications of being a carrier, why the father has to be tested, implications for the baby and implications for other children.

Table 3 Whether women were asked for their consent for thalassaemia carrier testing

	Groups			
Asked for consent to thalassaemia carrier testing?	Carriers ($n=19$)	Possible carriers ($n=17$)	Non-carriers ($n=110$)	Total ($n=146$)
Yes	—	3	7	10
No	14	14	101	129
Do not know/cannot remember	5	—	2	7

Barriers to acquiring information

Many women said that they did not know enough about the condition and/or test to ask health professionals any questions:

> *Fatima* (non-carrier, interviewed in English – 25 years old, primigravida, born in the UK): 'I just go to see her [midwife], but I don't ask many questions… but it's my first time yeah, I don't really know the questions.'

In addition, some women said that they did not ask for information because they believed that health professionals would automatically provide it for important tests.

> *Fozia* (non-carrier, interviewed in English – 20 years old, primigravida, born in the UK): 'I didn't [ask about tests] because I mean I thought well, you know, it's my midwife and if it was something important, maybe she would tell me herself and that I wouldn't need to ask.'

Most women said that they did not know which tests were important and so left the decision of providing information about important tests to health professionals.

Some of the women who were unable to speak English compromised their information requirements. They perceived not being able to speak in English as an obstacle placed by them in the process of communication. So, instead of expressing a desire for information, they accepted that they could not receive or obtain information:

> *Zora* (possible carrier, interviewed in Mirpuri – 38 years old, multigravida, 10 years in the UK): '…I don't speak or understand English properly. This is *my problem* [her emphasis]. I did ask the midwife why she was taking five bottles of blood. She did say something, but I don't know what because I didn't understand.'

However, given a choice, non-English-speaking women would have preferred some pre-test information:

> *Lubna* (possible carrier, interviewed in Mirpuri – 19 years old, multigravida, four years in the UK): 'Yes [would want information in own language]… it's obvious that one can then do everything for oneself. If you understand, then you could ask questions…'

The following quote shows how difficult it was for a non-English-speaking woman to obtain information, even though she had a strong need and desire for it:

> *Abida* (possible carrier, interviewed in Mirpuri – 30 years old, multigravida, 10 years in the UK): '… I'm having twins …I don't speak English. My husband usually goes with me, but he never explains anything to me properly. They explain everything to him, but he only tells me a little of what he's told. I want things to be explained to me … I really want to know what's going on… I want to know more. I want to ask more.'

If it is so difficult to ask for information when it is desired and needed, then it is likely to be more difficult when women do not know what tests are being performed or what questions to ask. The above quote also shows that health professionals should not assume that an individual will receive sufficient information, satisfactorily, via a relative who interprets for them, or that individuals would be able to ask for information they require via such an interpreter:

> *Jameela* (β-thalassaemia carrier, interviewed in Urdu – 22 years old, multigravida, two years in the UK): 'So when the midwife came, I said to my sister-in-law that "she is taking my blood so ask her about thalassaemia". She [sister-in-law] said that "if there is [thalassaemia] then she will tell you herself", so I said "okay".'

Jameela did not know that she had been tested until she received her positive test results.

Another reason women may not ask for information is because they believe that there is no need since their antenatal notes, which they keep and take to antenatal clinics, will be able to tell them what tests they have had. For example, a non-carrier interviewed immediately after receiving her routine antenatal test results looked through her notes to find out whether she had been tested for thalassaemia carrier status. Her notes showed that she had had the 'haemoglobinopathy screen' and the result was 'sickle cell screen – negative' and 'no abnormal haemoglobins', but thalassaemia was not specifically mentioned. So while some women believed their notes to be a source of information, they were unable to extract information about thalassaemia carrier testing because they did not understand what was written. These findings suggest the need for clearer reporting of negative test results.

Consent for antenatal thalassaemia carrier testing

In response to a question in the questionnaire, 129/146 (88.4%) of the women said that they were not asked for their consent for thalassaemia carrier testing (Table 3).

Attitude towards consent for thalassaemia carrier testing

Most women who knew that they had been tested believed that thalassaemia carrier testing was routine and that they had no choice about testing. Some women believed that if they had the right to choose, their midwife would have asked them. However, many women acknowledged that they had insufficient knowledge about various conditions or tests to make decisions about which ones they should be tested for:

> *Yasmin* (β-thalassaemia carrier, interviewed in English – 22 years old, primigravida, born in the UK): 'They [women] don't know what thalassaemia is and whether they should be having a test for it, but the doctors do…'

Many women viewed doctors and midwives as trained experts, who were competent in caring for mothers and babies. They suggested that because health professionals 'know best' and because women had comparatively little or no knowledge of the condition, the health professionals, rather than the pregnant woman, should decide which conditions she should be tested for:

> *Haleema* (β-thalassaemia carrier, interviewed in English – 19 years old, primigravida, born in the UK): 'No I don't think you can pick and choose [which tests you have]. What would women know about, you know, the doctor knows more…'

Another reason women allowed health professionals to carry out various tests without understanding much about them was because they trusted health professionals:

> *Sadia* (non-carrier, interviewed in Punjabi – 22 years old, primigravida, seven months in the UK): '…obviously whatever tests they do, they would think that it is better for us… If they think it is better to do these tests, then it's okay, let them.'

Women perceived doctors and midwives in a caring role, as well as in a position of power and authority, and therefore placed themselves in the health professionals' hands, entrusting them to do whatever they thought was best:

> *Interviewer*: 'So do you think that the way in which they tested you [without any information] was right or not?'
> *Jameela* (β-thalassaemia carrier, interviewed in Urdu – 22 years old, multigravida, two years in the UK): 'I think that they have done very well to tell me (that I am a carrier). At that time I thought "how good these people are, that they care about us so much [kithna khayal rakhte hein], because it's obvious they did it for us…"'

Three women were unhappy about being tested for thalassaemia carrier status without having given prior consent – all were articulate and professional women and knew that they had been tested before meeting the researcher. Only one non-carrier was genuinely concerned about consenting to antenatal thalassaemia carrier testing, and even this was because she worked in the legal field and was aware of her right to consent to testing. Furthermore, only two of these women said that they would have refused thalassaemia carrier testing. In one of these cases, the woman was already aware that she was a carrier and that her husband was not. She was angered about being re-tested because she had specifically agreed with her midwife not to be tested again and believed re-testing to be a waste of her own and health professionals' time and resources. In the other case, the possible carrier (in her third pregnancy) revealed that she only became concerned about not having given prior consent to testing when she realized that the test had been done as a matter of routine, and not at her midwife's request.

> *Shazana* (Possible carrier, interviewed in English – 35 years old, multigravida, born in the UK): '…when I went back to her [midwife], she was the one that said to me "oh, I didn't know you'd been tested for it." …that made me feel even worse'.'

Importance of testing versus consent

Many women also suggested that it was better for health professionals to test for thalassaemia carrier status without obtaining their consent prior to testing, than not to test at all:

> *Zakia* (β-thalassaemia carrier, interviewed in English – 23 years old, primigravida, born in the UK): 'I mean if they know it's quite common and they test you anyway [without asking], at least you know you've got it or you're carrying it. I wouldn't have known just to go and get myself tested for it. And if they've done it, fair enough, at least I know, it's better than not knowing…'
> *Interviewer*: 'Do you think they should ask?'
> *Zakia*: 'I don't know. It doesn't bother me though. It's something that I need to know.'

Many of the women felt that it was important to know whether there was anything wrong with them or their baby. Therefore, women approved of health professionals testing them for whatever conditions they considered to be important, without obtaining prior consent. One carrier thought that antenatal testing for thalassaemia was so important that she suggested that women should not be offered the choice of whether they wanted thalassaemia carrier testing because they may refuse to have it through lack of understanding about the importance of the test.

DISCUSSION

In view of the NHS Plan for commitment to a national antenatal and neonatal screening programme for thalassaemia and sickle cell in 2004, this is a timely study which aimed to explore the issues of informed consent and perceived pre-test information needs within a pregnant population. The strengths of this study are that (a) it is the first to explore attitudes towards informed consent for antenatal thalassaemia carrier testing, (b) the sample includes women with a variety of experiences of antenatal carrier testing and (c) it includes data from Pakistani women regardless of literacy or ability to communicate in English.

The present study, like a number of others,[22–25] has shown that there is a gap between the information women want about tests during pregnancy and the information provided by health professionals. Although the data in the present study were collected from women an average of two months after the event, and therefore could reflect faulty recall, they accord with observational studies[26–28] and support the pattern of results reported elsewhere,[29] including a Health Technology Assessment review on 'Psychosocial aspects of genetic screening of pregnant women and newborns', which very clearly shows that lack of understanding within this context is widespread.[30]

The findings also show that non-English-speaking women refrained from requesting information in a different language. There may be a number of reasons for this. One reason is that assertiveness in a medical context is culturally perceived as inappropriate, and women did not want to be seen as lacking confidence in their midwife/doctor or as questioning their authority by asking for information. Another reason for not asking questions may be the fear of being labelled as troublemakers.[31,25] Women may feel that they are dependent on health professionals for antenatal care, childbirth and postnatal care, and are likely to want to avoid being labelled a troublemaker by refraining from asking for information in a different language. In addition, women may not want to be seen as 'special needs cases', or as creating extra work for their midwife. During the interviews the researcher observed that most women were reluctant to say anything that could be interpreted as critical of midwives or antenatal services. The researcher felt that women genuinely appreciated the antenatal services available 'to the extent that they did not feel that they had

the right to hold, let alone state, their individual preferences'.[31] Like others,[19] the present study found that even if women know their information needs, they did not always know how to communicate them. Overall, women displayed compliant behaviour, where desire for information came second to building up and/or maintaining a good relationship with the midwives. This should not be interpreted as women not wanting information, just that they did not feel they could ask for it.

The main aim of the study was to explore pregnant women's views about informed consent. The findings showed that the concept had little meaning for these women. Instead, women talked about 'information' and 'consent' as separate issues, where most of them wanted to be informed about testing but did not necessarily want to consent, partly because of their views on who should make such decisions. That is, many of the women said that they trusted doctors and midwives because of their perceived expertise and competence in caring for mothers and babies, and believed that since health professionals 'know best' then they, rather than pregnant women, should decide which antenatal tests should be carried out.

While most women wanted to know that they were being tested for thalassaemia carrier status, they did not expect their consent to be obtained prior to testing. This may be because women were not aware of a right to choose to have or not have thalassaemia carrier testing. Like other routine antenatal tests, they did not expect to be involved in decision-making about whether or not they should be tested. Furthermore, the findings show that most women did not want to make decisions about antenatal thalassaemia carrier testing: they preferred to leave such decisions to the health professionals instead. This could be because they had poor knowledge about the conditions being tested for, and also because of the trust they had in their health professional as the decision-maker.[32] Others suggest that women believe in the knowledge and competence of health professionals, placing them in a position of authority and allowing them to make decisions about health care.[19] However, some of the women in the present study stated that they would have preferred to be given choices about antenatal tests and would have wanted to make decisions themselves. Therefore, health professionals could provide basic information about the conditions for which antenatal testing is being offered and determine the extent to which women want to be involved in decisions about which tests to have.

The majority of women showed that they were not concerned about giving consent for antenatal thalassaemia carrier testing. However, it is not clear how they would feel about consent for antenatal testing for other disorders, such as cystic fibrosis, Down's syndrome or late-onset conditions.

Overall, there are a number of implications for service provision. Schemes such as 'Informed Consent for Antenatal and Neonatal Screening' (commissioned by the National Screening Committee) and 'PEGASUS' (a training network commissioned by the NHS Sickle Cell and Thalassaemia Screening Program) to develop training for health professionals to facilitate the implementation of the Program, with a focus on training for frontline health professionals (such as midwives and GPs), are currently in the pipeline. Our findings show that such training should include information on laboratory investigations for thalassaemia carrier status, showing how the first stage of this investigation involves a routine MCH measure that is performed for all women. This would enable frontline health professionals to understand how and why women could be tested for thalassaemia carrier status even if they do not request such testing. Furthermore, there is a need for guidelines that allow laboratories to identify women who have not consented to testing following the initial routine MCH screening test, and then specify how to obtain consent so that women who have not already been approached can opt out of further testing at this stage if they wish to do so.

In relation to information provision and people's needs for varying amounts of information, one way forward could be for health professionals to assess pre-test information needs on an individual basis and provide information accordingly. However, there are problems with this because of the time constraints on health professionals. An alternative is to provide information in written form (i.e. leaflets in different languages)[33] so that women can extract information according to their own needs.

Leaflets about being a thalassaemia carrier and how thalassaemia is inherited are currently available, which have been developed by the UK Thalassaemia Society[34] and by individual health authorities. However, the language used in both the English and particularly the Urdu version is complicated and sometimes misleading. For example, in their review of genetic information available in Urdu, Shaw and Ahmed[33] found inaccuracies in leaflets differentiating carriers from affected individuals. There is a need for a leaflet specifically designed for antenatal thalassaemia carrier testing (initially in English and then in other languages). Such a leaflet should be evaluated for its ease of accessibility, its ability to meet women's perceived pre-test information needs and its ability to serve as a reference point for women receiving positive results. Perhaps separate leaflets should be developed for β-thalassaemia and α-thalassaemia carriers in order to reduce the confusion that could arise from presenting the two in the same leaflet. Women with information needs exceeding those provided in the leaflet could be referred to specialists, such as thalassaemia counsellors, instead of expecting midwives to provide detailed information. Of course, leaflets are only useful if women can read them. For women unable to read, yet another alternative would be to provide information in the form of audio or videotapes.

CONCLUSION

The analysis has allowed the deconstruction of the concept of informed consent for these women. The findings show that while information was important to women, consenting was not. Overall, women discussed information and consent as two separate issues, thus challenging many of the assumptions around the term informed consent. They wanted pre-test information because they just wanted to know, not to use it to make decisions about testing.

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