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Ethnicity Questions and Antenatal Screening for Sickle Cell/Thalassaemia [EQUANS] in England: A Randomised Controlled Trial of Two Questionnaires

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Concepts allied to ethnicity are increasingly coming under question as legitimate variables for use in health research. A randomised controlled trial of two ethnicity screening questions for ascertaining risk of carrying genes associated with sickle cell and thalassaemia illustrates the challenges and limitations of assessing an association of social constructs and genetic statuses.

Objectives. *To evaluate two candidate ethnicity screening questions in antenatal screening programmes in low, mixed and high sickle cell prevalence areas, and to identify time taken in administration of the questions by use of the following measures:*

(1) Proportions of respondents with missing ethnicity data and/or significant changes in ethnic/family origins upon re-interview.
(2) Numbers of carriers of clinically significant haemoglobin disorders missed by ethnicity screening questions.
(3) Time taken to explain screening question for sickle cell disease (SCD)/thalassaemia and obtain ethnic/family origins.
(4) Proportion of clients providing usable ethnic/family origins data.
(5) Reported ethnic/family origins in pregnant women at first booking with midwife.

Design. *Ten-month (September 2002–June 2003) questionnaire study with random allocation to two self-administered ethnicity questions, comparison with laboratory results and results from re-interview. The settings were antenatal booking clinics in four geographical areas of England of varying expected foetal prevalence of SCD: very high*

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(29.75 per 10,000 pregnancies); high (8.2); mixed high and low (1.29); and low (0.18). The subjects were 4,559 pregnant women at first booking with midwife.

Results. *Proportions of respondents with missing ethnicity data and/or significant changes in ethnic/family origins upon re-interview were 4.33% (CI 2.63–6.68%) for a category-based question and 9.45% (CI 6.86–12.61%) for a binary plus open-ended question. Proportions of carriers missed were 5.74% (CI 2.34–11.46%) and 9.71% (CI 4.75–17.13%) by category-based and binary plus open-ended questions, respectively. Average time taken to ascertain ethnic/family origins for screening was between 2.17 and 5.12 minutes in different areas, and up to 15 minutes at the 95th centile. Usable ethnicity screening data was missing in 2.94% of instances. Errors in interpretation or missing data were 3.2% for a category-based question and 4.71% for a binary plus open-ended ethnicity question. Ethnicity Question A produces fewer cases of missing or misinterpreted data ($p < 0.001$).*

Conclusions. *A category-based ethnicity screening question was more effective than a binary plus open-ended question. Using the more effective question, 5.74% (CI 2.34–11.46%) of significant haemoglobinopathies will be missed in a selective screening programme, and 4.33% (CI 2.63–6.68%) of replies to an ethnicity screening question will be unreliable when compared to information given upon re-interview. In specific carefully circumscribed situations, namely, in antenatal screening for sickle cell and thalassaemia, it is possible to measure the degree of association between social constructs of ethnicity and health status in a manner that may help in effecting policy decisions.*

Keywords: Ethnicity; Sickle Cell; Thalassaemia; Screening

Introduction

In this paper, we take the evolving debate on the relationship between ethnicity and health as a viable concept for health research and consider it in the light of applied health research focusing on antenatal screening for sickle cell and thalassaemia. We begin by outlining some of the ongoing debates concerning the place of a concept such as ethnicity in health research; discuss the background to antenatal screening for sickle cell and thalassaemia in England; and report on the results of a randomised controlled trial of two different ethnicity screening questions designed to help target laboratory testing for sickle cell, other haemoglobin variants and for the thalassaemias. Finally, we assess the implications for ethnicity and health research of the results of the trial of the two questionnaires.

The Concept of Ethnicity and Health Research

Sheldon and Parker (1992) were the first to draw attention in principle to the dangers of reifying the concept of ethnicity in health research, dangers empirically illustrated

by Nazroo (1999). McKenzie and Crowcroft (1996) attempted to deal with the issue by urging the comprehensive, some might say indiscriminate, collection of data and a full description of what is then found, but Bradby (2003) questions the universal applicability of the concept of ethnicity for health research. Ellison (2005) suggests that the race/ethnicity concept should be predominantly restricted to assessing the impact of discrimination, and that only under limited specified conditions should it be used as a proxy for an associated health factor.

To this extent, a rejection of a category as a universal entity does not necessarily imply that a concept allied to ethnicity (following authors such as Berthoud 1998, 'ethnic/family origins' is the term used as part of the ethnicity questions trialled in this study), a concept devised with a specific rather than a universal purpose in mind, may not have some utility for situations where the parameters are carefully drawn, and the specific rationale for the question explained to the client. As Hindess (1973) has argued, demonstrating the variability in meaning of a concept does not tell us how frequent that variation is, nor the practical consequences of that variation. Developing an ethnicity question based on theoretical work in constructing categories best suited for the specific purpose of effecting selective screening for sickle cell and thalassaemia could result in relatively few carriers being missed because of ambiguities in the question categories themselves. However, in routine antenatal screening practice, factors such as the pressure of workload; the tendency of health professionals to circumvent asking the question; and the possibility that the act of asking an ethnicity question could reinforce false notions of distinct biological 'races' could simultaneously reduce the effectiveness of the ethnicity questions devised as valid and reliable screening tools.

Sickle Cell and Thalassaemia Screening

Sickle cell disease (SCD) and the thalassaemias are inherited haemoglobin disorders that mainly affect people of African, Caribbean, Middle Eastern, South Asian, South East Asian and Mediterranean descent (Serjeant & Serjeant 2001; Weatherall & Clegg 2001), but are also rarely found in the Northern European population (Lehman & Huntsman 1974). In general, policy makers in the UK have failed to see sickle cell and thalassaemia as issues affecting *all* populations, and, in contrast to conditions mainly affecting majority ethnic populations in England (such as cystic fibrosis and haemophilia), sickle cell and thalassaemia have, until recently, seen the persistence of poorly resourced and uneven services (Atkin & Anionwu 2001), including screening services (Atkin & Ahmad 1998; Atkin *et al*. 1998).

For the NHS Sickle Cell and Thalassaemia Screening Programme in England, one purpose of antenatal screening is to identify and facilitate informed reproductive choices to women and couples identified as being at higher risk of carrying genes associated with sickle cell or thalassaemia (NHS Sickle Cell and Thalassaemia Screening Programme 2004). Classic international advice on screening suggests that priority should be accorded to informing 'well-defined populations' (Lappé *et al*.

1972, p. 1130). There is a strong, though gradually dissociating, relationship between socially constructed categories such as ethnicity and risk of carrying genes associated with SCD/thalassaemia (Department of Health 1993; Andrews *et al*. 1994). Determination of ethnicity has been seen for many years in England as part of the antenatal screening process for sickle cell, the thalassaemias and other haemoglobinopathies (Department of Health 1993), and, although this has been widely practised (Sedgwick & Streetly 2001), it is not known how best ethnicity should be determined for this specific purpose, nor how reliable this screening process is. Assessing the validity and reliability of ethnicity screening questions can contribute to policy decision-making regarding the relative merits of selective and universal antenatal screening for sickle cell and thalassaemia (Zeuner *et al*. 1999; Davies *et al*. 2000).

There are, however, very real challenges in using a social construct such as ethnicity to define populations at risk by virtue of genetic lineage (Dyson 1998, 1999, 2005). Although evidence-based rates for ethnic-specific prevalence have been produced for England (Hickman *et al*. 1999; Davies *et al*. 2000), there has been a wide and conceptually inconsistent range of ethnic categories in use for ethnic ascertainment in antenatal clinics in England (Bain & Chapman 1998; Aspinall & Dyson 2002). It was against this background that the National Health Service Plan for England promised '. . . a new national linked antenatal and neonatal screening programme for haemoglobinopathy and sickle cell disease by 2004' (Department of Health 2000, p. 13.16).

In the UK, most areas use screening algorithms which rely on ethnic information to determine the risk of someone carrying a haemoglobinopathy ('high risk if any ethnic history outside of UK or if ethnic/family origins uncertain/unknown'—NHS Sickle Cell and Thalassaemia Screening Programme 2005a, p. 1), before deciding which laboratory test should be offered (*selective* screening). Areas designated as having a high prevalence of haemoglobinopathies practice *universal* antenatal screening, in which full laboratory testing (full blood count and high performance liquid chromatography (HPLC) to identify any haemoglobin variants) is offered to everyone regardless of their apparent ethnic group (universal screening). Although selective screening in areas of low haemoglobinopathy prevalence is potentially more cost-effective, it critically relies on being able to determine ethnic origin in a reliable fashion. Accurate ethnic information is also important in universal screening programmes for the interpretation of results of full blood counts, particularly in determining the risk of severe alpha-thalassaemia. A mean cell haemoglobin result of less than 25 pg is regarded as 'high risk if any ethnic/family origins in China (including Hong Kong), Thailand, Taiwan, Cambodia, Laos, Vietnam, Indonesia, Burma, Malaysia, Brunei, Singapore, Philippines, Cyprus, Greece, Turkey or if ethnic/family origins uncertain/unknown' (NHS Sickle Cell and Thalassaemia Screening Programme 2005a, p. 2).

In summary, there is little evidence to suggest how ethnicity information for haemoglobinopathy screening should be determined (Streetly 2000), particularly in

low prevalence areas where health staff are less comfortable in seeking ethnic data and take longer to collect it (Pringle & Rothera 1996). In this paper we report on an assessment of the relative merits of two ethnicity questions. In order to determine the respective strengths of the questions, a number of key factors in the performance of the questions need to be examined. First, we need to know the extent to which each ethnicity question is valid. Second, a requirement of a screening question is that it is reliable. Third, the time taken by the midwife to ask the ethnicity question for the purposes of effecting antenatal screening for sickle cell/thalassaemia needs to be noted, so that policy makers may account for the full economic costs of a screening programme.

Methods

The objectives of the study were to compare two different ethnic questions for stability and for proportion of carrier mothers[1] missed, whilst assessing the overall feasibility of determining ethnic group with reference to haemoglobinopathy screening. Candidate ethnicity questions, previously designed (Aspinall & Dyson 2002), were further developed through (1) reducing the three candidate questions to two by means of a pilot study with 330 university students and 62 health professionals that assessed the acceptability of the respective questions; (2) discussions with midwifery teams administering the ethnicity screening questions in antenatal settings to assess the feasibility of midwives completing the research instrument, which led to a reduction in the number of questions within the research instrument; (3) the addition to both questions of the same introductory paragraph, explaining the sickle cell and thalassaemia-related reason for asking the question; (4) incorporating the amended questions into a 10-page research instrument; (5) piloting of the research instrument for one month with 30 mothers in one routine antenatal practice setting.

Ethnicity Question A is a classification question similar in structure to the 2001 Census England and Wales question (Figure 1). It also contains extra categories to capture all appropriate haemoglobinopathy risk groups and a 'tick all that apply' method (as opposed to categories) to record mixed heritage (Aspinall *et al*. 2003). Ethnicity Question B comprises two parts (Figure 2). Part One contains an initial binary question to identify those with ancestors outside of the UK/Eire. This is followed by Part Two, free text provision to write in countries of ethnic/family origin, with up to five free text boxes supplied in order to capture mixed heritage (Aspinall & Dyson 2002).

In order to answer the research questions concerning the respective validity and reliability of the candidate ethnicity questions, and the administrative burden in time represented by asking such questions in routine practice, the research has three principal outcome measures. First, we have assessed the validity of the respective ethnicity questions by offering a laboratory screen to all women, identifying carriers by laboratory means, and then assessing how many carriers would have been missed

We are asking about ethnicity because we want to know who is at risk from sickle cell/thalassaemia. These are serious inherited blood disorders that are more common in peoples whose ancestors lived in malarial areas of the world such as Africa, Asia, the Middle East, and the Mediterranean. Bearing this in mind …

DO YOU HAVE ETHNIC/FAMILY ORIGINS THAT ARE …

Please tick one or more boxes to indicate these origins

A. WHITE
- English, Scottish, Welsh, or Irish □
- Other North European □
- Greek or Greek Cypriot □
- Turkish or Turkish Cypriot □
- Italian, Maltese, or other Mediterranean □
- Any other White background (*please write in.*……) □

B. MIXED ► *Please tick all boxes in sections A, C, D, and E (above & below) that apply to you*

C. ASIAN OR ASIAN BRITISH
- Indian or African-Indian □
- Pakistani □
- Bangladeshi □
- Any other Asian background (*please write in.*……) □

D. BLACK OR BLACK BRITISH
- Caribbean □
- African □
- Any other Black background (*please write in.*……) □

E. CHINESE AND OTHER
- Chinese □
- Japanese □
- Malaysian, Vietnamese, or Filipino □
- North African, Arab, or Iranian □
- Any other (*please write in.*……) □

Ethnicity Information Refused □

Figure 1 Ethnicity Question A. *Source*: Developed from Aspinall *et al*. (2003).

had the ethnicity screening questions been relied upon to select those at risk. Second, we have judged the reliability of ethnicity questions by asking the ethnicity question at the time the mother first visits the midwife to 'book' her pregnancy, when the question is asked by the midwife, and then a different person asking the mother the same ethnicity question at a subsequent antenatal visit several weeks later. This is because if the ethnic self-ascertainment is found to depend either upon timing or upon any difference in the identity of the interviewer, then to that extent that particular ethnicity question is unreliable for the purposes of constituting a screening tool. In other words, a screening question must be sufficiently robust not to be affected by any (inevitable) interviewer bias in the real life setting of routine health

We are asking about ethnicity because we want to know who is at risk from sickle cell/thalassaemia. These are serious inherited blood disorders that are more common in peoples whose ancestors lived in malarial areas of the world such as Africa, Asia, the Middle East, and the Mediterranean. Bearing this in mind …

1. Do you or any of your known ancestors, as far back as you can recall, have ethnic/family origins from areas of the world outside of the United Kingdom or Republic of Ireland?

Please tick one box only.

Yes □
No □
Don't Know □

2. If Yes, then for you or for any of your known ancestors, as far back as you can recall, please write in all the countries in the spaces below:

Ethnicity Information Refused □

Figure 2 Ethnicity Question B. *Source*: Developed from Aspinall and Dyson (2002).

care practice. Third, we have measured the time taken by the midwife to ask the ethnicity question for the purposes of effecting selective screening for sickle cell/thalassaemia, noted.

The study was conducted in four hospital trusts with widely varying prevalence of minority ethnic groups and haemoglobinopathies. The areas were a very high prevalence area (expected foetal prevalence of sickle cell disease (SCD) 29.75 per 10,000); a high prevalence area (8.2); an area of mixed high and low prevalence (1.29); and a low prevalence area (0.18). Fifty-seven half-day preparatory workshops were held with 262 community midwives in the four study areas in order to prepare them for their role in data collection.

The study was approved by a Multi-Centre Research Ethics Committee and by the four Local Research Ethics Committees. Questionnaires containing one of the two ethnicity screening questions were sealed in opaque envelopes and placed in random order by use of a random number table before distribution to screening midwives. All pregnant women presenting to a midwife for their first antenatal appointment (the 'booking-in interview') during the study period were eligible for inclusion in the study. Neither midwife nor mother knew the allocation to question before the opening of the opaque envelope, which occurred after consent was obtained. Consenting clients were asked one of these two randomly assigned ethnicity questions and (except in the low prevalence area) were offered a haemoglobinopathy test by standard laboratory methods (British Society for Haematology 1988), including full blood counts and haemoglobin HPLC to identify HbS, HbC, HbD, HbE, HbO Arab and beta-thalassaemia carriers. After four months of the study, the design was amended to permit the recruitment of carriers (not previously recruited by the midwife) at the later point of contact with the haemoglobinopathy counsellor. For these clients, ethnic self-ascertainment was influenced by the fact they were asked for the information following receipt of a carrier result. The data were collected using a questionnaire in three sections: ethnicity information (self-administered), laboratory results and ethnicity information at re-interview. The re-interview was carried out on a subsequent occasion (for example, at the next antenatal visit). Re-interviews were conducted with a one in 10 systematic sample[2] of all women in the study; with all those identified as carriers of a significant haemoglobinopathy, and with those with borderline results. In the re-interview, the woman was asked to assign her ethnic/family origins on a second occasion and this result was compared with the original answer (Figure 3). The power to detect significant differences upon re-test between Question A and Question B at the 5% significance level with sample sizes of approximately 436 was 85%. All data were analysed using SAS®, and results are presented with 95% confidence intervals.

Results

Level of Recording of Ethnic Data

A total of 4,559 women consented to take part in this study (out of 5,211 recorded as invited from a total of 19,546 undergoing antenatal booking during this period). Checks with the recruiting midwives confirmed that an overwhelming majority of those women eligible for the study were never invited to participate, as opposed to being asked but not recorded as declining. 2,316 answers were received for Ethnicity Question A and 2,250 for Ethnicity Question B. In none of the areas did the capture of ethnic data for the purposes of selective antenatal screening for sickle cell/thalassaemia achieve the levels of coverage for standard ethnic monitoring data for that area (Table 1). Of the women recruited to the study, ethnicity screening information was missing for 2.94% (134/4,559).

The Ethnicity Questions

The ethnic/family origins data captured by Ethnicity Questions A and B are given in Tables 2–4. Responses to Part Two of Ethnicity Question B have been mapped to the categories derived from Ethnicity Question A (see Table 2) based on the client's responses. This inevitably involves judgements about what category a respondent may have used.

For Ethnicity Question A, the total number of cases of missing data, or uninterpretable data amounted to 33 out of 2,313 answers (3.2%). On Part One of Ethnicity Question B, the ethnicity data was refused, missing or ambiguous in 13/2,247 cases (0.58%). In addition, there were 86 *Don't Know* responses. Part Two of Ethnicity Question B also produced missing data and misinterpretations in 34/823 (4.13%) instances. The combined error rate for both parts of Ethnicity Question B is 4.71%.

Table 1 Comparison of Level of Ethnicity Data Capture, Standard Ethnic Data and EQUANS Ethnicity Questions, All Areas

Area	Data	Ethnic monitoring data, antenatal population	Ethnicity data collected, EQUANS study		
			Compared to whole antenatal population	Compared to those recorded as invited into the study	Compared to those recruited to the study
Very high prevalence	Collected	4,108	1,442	1,442	1,442
	Missing	6 [0.15%]	2,671 [64.9%]	275 [16.0%]	36 [2.4%]
	Total	4,114	4,113	1,717	1,478
High prevalence	Collected	2,906	152	152	152
	Missing	102 [3.4%]	2,856 [95.0%]	87 [36.4%]	7 [4.4%]
	Total	3,008	3,008	239	159
Mixed prevalence	Collected	9,190	1,953	1,953	1,953
	Missing[a]	92 [1%]	7,329 [79.0%]	241 [11.0%][b]	74 [3.7%]
	Missing[c]	1,354 [14.6%]			
	Total	9,282	9,282	2,194	2,027
Low prevalence	Collected	3,104	878	878	878
	Missing	38 [1.2%]	2,264 [72.1%]	183 [17.2%]	17 [1.9%]
	Total	3,142	3,142	1,061	895

[a]Excludes one entire unit unable to provide any ethnicity data.
[b]Includes two who declined to be screened, but who completed ethnicity data.
[c]Includes figures from an entire unit unable to provide any ethnicity data.

Table 2 Responses to Ethnicity Question A, 'Do You Have Ethnic/Family Origins That Are . . .'

	Very high prevalence	High prevalence	Mixed prevalence	Low prevalence
WHITE				
English/Scottish/Welsh/Irish	235	9	727	382
Other Northern European	21	1	3	3
Greek/Greek Cypriot[a]	6	0	2	0
Turkish/Turkish Cypriot[a]	8	0	1	0
Italian, Maltese or Mediterranean[a]	7	0	8	5
Any other white background	32	1	14	4
Mixed white—not at risk	19	0	21	12
Mixed white—at risk[a]	21	0	28	12
MIXED[b]	4[c]			
Mixed MEG and white[a]	53	11	29	15
Mixed MEG only[a]	21	2	4	1
ASIAN OR ASIAN BRITISH				
Indian or African-Indian[a]	22	13	99	2
Pakistani[a]	4	24	13	0
Bangladeshi[a]	9	3	6	0
Any other Asian background[a]	7	2	9	1
BLACK OR BLACK BRITISH				
Caribbean[a]	100	17	7	0
African[a]	153	3	21	0
Any other black background[a]	8	0	4	0
CHINESE AND OTHER				
Chinese[a]	4	0	2	0
Japanese[a]	1	0	1	0
Malaysian, Vietnamese or Filipino[a]	1	0	3	3
N. African, Arab or Iranian[a]	3	0	0	1
Other origins[a]	5	1	5	0
MISINTERPRETATIONS	4[d]	0	4[e]	2[e]
MISSING DATA	4	1	19	1
PAGE IN PROFORMA MISSING	0	0	1	0
TOTAL	750	88	1,031	444

[a]Origins previously deemed at risk, or possibly at risk, of carrying genes associated with sickle cell/thalassaemia.
[b]Involving at least one minority ethnic group (MEG).
[c]Indicated mixed, ticked no boxes.
[d]Comment about partner, no category given.
[e]Ticked white (English), annotation(s) by midwife suggested the wrong category ticked.

Table 3 Response Options to Ethnicity Question B (Part 1: Do You or Any of Your Known Ancestors, as Far Back as You Can Recall, have Ethnic/Family Origins from Areas of the World Outside of the United Kingdom or Republic of Ireland?)

	Very high prevalence	High prevalence	Mixed prevalence	Low prevalence
Yes	429[a]	47	259[b]	72[c]
No	265[d]	18[d]	694[e]	364[d]
Don't know	29[f]	6	36[g]	15[h]
Info refused	0	0	2	0
Blank	3	0	4	0
Ambiguous	3[i]	0	1[j]	0
Total	729	71	996	451
Error rate			Blanks + ambiguous responses/total 13 out of 2,247 = 0.58%	
Error rate + don't know			Don't knows + blanks + ambiguous responses/total 99 out of 2,247 = 4.41%	

[a]Includes two where white (English/Scottish/Welsh/Irish) ethnicity was given for Yes.
[b]Includes two where white (English/Scottish/Welsh/Irish) ethnicity was given for Yes.
[c]Includes two where white (English/Scottish/Welsh/Irish) ethnicity was given for Yes and three missing ethnicity.
[d]Includes one where high risk ethnicity was given for No.
[e]Includes four where high risk ethnicity was given for No.
[f]Includes one where high risk ethnicity was given for Don't Know.
[g]Includes three where high risk ethnicity was given for Don't Know.
[h]Includes two where high risk ethnicity was given for Don't Know.
[i]Includes two who ticked Yes and Don't Know (both then wrote high risk countries) and one who ticked No and Don't Know who then gave no further information.
[j]Includes one who ticked Yes and No and then wrote a high risk country.

Test/Re-test Reliability

A test/re-test compared ethnic ascertainment at first antenatal booking and upon re-interview. These re-tests suggest the extent to which an ethnicity screening question may prove unreliable in practice. Errors were held to comprise occasions when a respondent gave a significantly different answer than before (that is, moving them from risk to non-risk categories for haemoglobinopathies or vice versa) or no usable answer on one of the two occasions. On the test/re-test component of the study Question A performed better than Question B. The overall error rate for reliability for Question A is 19/439 (4.33%, CI 2.63–6.68%), which is significantly different ($p = 0.003$, CI -8.5-1.8%) from the overall reliability for Question B, which is 41/434 (9.45%, CI 6.86–12.61%).

Table 4 Responses to Question B (Part Two: If Yes, then for You or for Any of Your Known Ancestors, as Far Back as You Can Recall, Please Write in all the Countries in the Spaces Below)

	Very high prevalence	High prevalence	Mixed prevalence	Low prevalence
WHITE				
English/Scottish/Welsh/Irish	2[a]	0	3[a]	2[a]
Other Northern European	36	0	24	28
Greek/Greek Cypriot[b]	5	0	0	1
Turkish/Turkish Cypriot[b]	0	0	3	0
Italian, Maltese, Mediterranean[b]	4	0	12	4
Any other white background	23	1	17	10
Mixed white—not at risk	4	0	3	1
Mixed white—at risk[b]	13	0	3	4
MIXED[d]				
Mixed MEG + white[b]	45	7	13	3
Mixed MEG only[b]	38	4	41	2
ASIAN OR ASIAN BRITISH				
Indian or African-Indian[b]	14	5	46	4
Pakistani[b]	3	14	4	0
Bangladeshi[b]	3	4	7	0
Any other Asian background[b]	5	1	5	3
BLACK OR BLACK BRITISH				
Caribbean[b]	84	8	15	3
African[b]	129	3	35	1
Any other black background[b]	0	0	1	0
CHINESE AND OTHER				
Chinese[b]	1	0	4	2
Japanese[b]	0	0	0	0
Malaysian, Vietnamese or Filipino[b]	2	0	5	0
N. African, Arab or Iranian[b]	7	1	5	0
Other origins[b]	6	0	4	4
MISINTERPRETATIONS[c]	2	0	3	2
MISSING DATA (TO 'YES')	9	0	17	1
TOTAL	433	48	267	75

[a]Represents a misinterpretation of the question.
[b]Origins previously deemed at risk, or possibly at risk, of carrying genes associated with sickle cell/thalassaemia.
[c]Included under white (English/Scottish/Welsh/Irish), above.
[d]Involving at least one minority ethnic group (MEG).

Comparison with Laboratory Results

225 carriers of clinically relevant haemoglobinopathies (defined as those in which haemoglobinopathy testing would be requested on the partner because of the risk of foetal haemoglobinopathy) and 249 with borderline laboratory results were reported. 122 carriers received Question A, 103 received Question B.

For Ethnicity Question A 7/122 (5.74%, CI 2.34–11.46%) carriers were missed. In two cases ethnic/family origins category at risk for haemoglobinopathies was only recorded at the time of the re-test interview. In these two missed cases the original form has other information completed, but the ethnicity data is missing. Three further cases recruited at counselling were never offered the ethnicity question at booking by the midwife, and are included as misses on the basis of intention to treat. One client declined the ethnicity question which was left blank but was found to be a carrier. One client ticked a low risk category, but had elevated foetal haemoglobin requiring partner testing. One further carrier ticked 'Any Other White Background', generally a low risk category, but wrote in 'Egypt', a high risk country, and has not been counted amongst those carriers deemed missed.

Ten out of 103 (9.71%, CI 4.75–17.13%) diagnoses of a significant haemoglobinopathy requiring partner testing were missed using Question B. In seven cases the client ticked 'No' in response to the question asking if they had ancestors with ethnic/family origins outside the UK/Eire but had a significant haemoglobinopathy on laboratory testing. In another seven cases, the client ticked 'Don't Know', though answering in this way could itself be regarded as a criterion for offering laboratory testing, and these have not been included in the cases deemed missed. In one further case the client declined the ethnicity question which was left blank, but was screened. Two further clients provided no indication of risk at the first interview, and only provided information on their risk group at re-test interview once their carrier status had been established.

Statistically, there is no significant difference between the validity values (proportions of carriers as measured by laboratory results compared to those identified as at risk by the respective ethnicity screening question) for the two questions ($p = 0.2615$ using a chi-square test), though there is a trend towards Ethnicity Question A performing better than Ethnicity Question B.

Timing

The mean time taken to ask the ethnic/family origin question in the four centres varied from 2.17 to 5.12 minutes (Table 5). The mean time taken to ask Ethnicity Question B (4.48 minutes, CI 4.47–4.69) was generally slightly longer than that for Question A (4.41, CI 4.19–4.63), except for the very high prevalence area where the pattern was reversed.

To summarise, for Ethnicity Question A, the sources of error as a screening question comprise the initial errors associated with the ethnicity question (3.2%),

Table 5 Time[a] Taken to Answer the Ethnicity Questions All Areas

	Very high prevalence	High prevalence	Mixed prevalence	Low prevalence
Mean (category question)	4.91	1.95	5.06	2.61
Mean (open response)	4.84	2.45	5.18	2.62
Mean (both questions)	4.87	2.17	5.12	2.61
95% Confidence level for mean	0.27	0.41	0.25	0.24
95% Centile	14.00	7.75	15.00	9.00
Standard error	0.14	0.21	0.13	0.12
Median	3.00	1.00	4.00	2.00
Mode	2.00	0.50	5.00	2.00
Standard deviation	4.94	2.61	5.61	3.32
Sample variance	24.40	6.81	31.50	11.05
Range	82.98	14.83	74.98	30.81
Minimum	0.02	0.17	0.02	0.02
Maximum	83.00	15.00	75.00	30.83
Count (valid cases)	1,313	153	1,907	822

[a]Times are cited in minutes, correct to two decimal places.

carriers identified by laboratory methods missed by the ethnicity screening question (5.74%) and the overall re-test error rate for that question (4.33%). For Ethnicity Question B, the errors identified are made up of the initial errors associated with the ethnicity question (4.71%), carriers missed by the ethnicity question (9.71%) and the overall re-test error rate for that question (9.45%).

Discussion

Test/Re-test Reliability

A widely cited, but ultimately arbitrary, maximum tolerable threshold for errors within a selective screening programme based on an ethnicity screening question is 5.5% (Zeuner *et al*. 1999). This threshold is met by the category-based question, but not binary plus open response question. The implication of this seems to be that whilst variability of response to an ethnicity question may be considerable, the type of variability liable to impact upon the specific, circumscribed purpose of the ethnicity screening question for ascertaining sickle cell and thalassaemia risk can at least be estimated, and policy decisions made on that basis.

Comparison with Laboratory Results

The maximum 5.5% level of carriers missed, a challenging level of service attainment modelled by a Health Technology Assessment Report for a 'poorly run' selective programme (Zeuner *et al*. 1999), is met neither by the category-based question, nor by the binary plus open response question. However, it is important to note that the

source of errors in Ethnicity Question A relates primarily (in six of the seven cases) to the administration (or non-administration) of the ethnicity screening question rather than to any conceptual ambiguity in the categories presented. By contrast, the source of errors in Ethnicity Question B is primarily associated with inadequacies inherent in the structure of the question, in that seven out of the ten missed carriers ticked 'No' in response to the question asking if they had ancestors with ethnic/family origins outside the UK/Eire.

Timing

The time taken to ask the ethnic questions varied widely, both within and between centres, and is in keeping with the variability in overall mean booking times, from 39 minutes in one high prevalence area, to 60 minutes in other areas. The high prevalence area had shorter times recorded for timing the ethnicity questions and, according to the local researcher who conducted observations of the bookings, this reflected the lack of explanation of sickle cell/thalassaemia as part of the greater rush to complete the overall booking within the given timeframe. In the low prevalence area the two local researchers reported the reluctance of a large majority of the midwives to provide *any* explanation of sickle cell/thalassaemia, and the midwives themselves reported that they were neither knowledgeable nor confident to do so. The figure for the low prevalence area may therefore reflect inappropriate practice in that little or no explanation was given.

This leads us to conclude that the timings upon which to base projections should be based on the two remaining areas with mean times of 4.87 and 5.12 minutes, respectively. If one were to take account of the pressures of routine practice and be 95% certain of providing the professional asking the screening question with sufficient time, the figure would be 14–15 minutes.

Level of Recording of Ethnic Data

A key area for further research would appear to be the efficacy of health staff in busy routine practice in actually asking the screening question in a comprehensive manner. This study only estimates the utility of ethnicity questions in people already recruited to a research study (4,559 of 5,211 or 87.5% of women reportedly invited in to the trial), but in practice a big additional source of error will arise from women not being asked the relevant ethnic question at all (in this case only 5,211 of 19,546 eligible women were invited in to the trial). In an additional arm of the study, not reported here, seven researchers observed and took contemporaneous notes on 121 booking-in interviews between mother and midwife; conducted 111 short taped interviews with the mother; carried out 115 short taped interviews with 61 different midwives; and kept fieldwork notes on 76 meetings conducted throughout the research process. On the basis of these data, the low attempted recruitment rate was reported by the midwives to be based on their busy schedules, and their own judgements about

whom to invite to be screened. This is likely to be a serious problem, and could significantly undermine the validity and reliability of an ethnicity screening question in routine health care practice, unless sufficient time is given during the antenatal booking process and the importance of haemoglobinopathy screening emphasised in education for delivery of antenatal services. At the moment we do not know to what extent the low level of coverage in asking the ethnicity question (23.3%), in a time-limited research project with full funding of midwifery time, would be mirrored in a permanent situation of standard service delivery.

Conclusion

A category-based ethnicity question was more reliable, missed fewer carriers and was slightly quicker to administer than an open-ended question. The category-based question had a re-test error rate of 4.33%, the open-ended question 9.45%. This category-based ethnicity screening question missed 5.74% of carriers (just outside the outer limit of what has been regarded as tolerable in a selective screening programme). An open-ended question missed 9.71% of carriers, outside the limits which could be countenanced in a selective programme. The time actually required for asking an ethnicity question, explaining sickle cell/thalassaemia and obtaining consent to screen is considerably higher than previous estimates. Administered correctly with accompanying explanation, asking an ethnicity screening question takes five minutes. To be 95% confident of providing health staff with sufficient time 14–15 minutes are required.

The universal utility of ethnicity as a concept for use in health research has been questioned (Bradby 2003). On the one hand, this is because ethnic categories are neither stable nor permanent classifications of peoples (Fenton 1999). Indeed, in referencing this notion of the fluidity of the concept ethnicity, various commentators have referred to situational ethnicities (Okamura 1981), plastic ethnicity (Gillborn 1995), ethnicities in the plural (Modood *et al*. 1994) or to processes of ethnic identification (Culley & Dyson 2001). Thus ethnicity can no longer be thought of as an essential quality residing deep at the heart of a person's identity. The particular aspect of a person's ethnic/family origins that they choose to foreground in any particular social situation may therefore be different. In principle, such commentators make a convincing case for the situational variability of a concept such as ethnicity, and therefore raise important questions about the very possibility of using ethnicity as a marker of genetic risk. However, it is possible, as this study shows, to make some estimates, not of how often this variability occurs per se, but at least how often the variability has consequences of not identifying real genetic risks in the manner intended by the screening programme.

On the other hand, the use of a category such as ethnic/family origins in the context of a genetic screening programme has the very real possibility of invoking at least cultural essentialism if not also a form of biological reductionism that assumes

(wrongly) the existence of distinct 'races' (Ellison 2005). For Ellison, therefore, the prime use of race/ethnicity concepts is to assess the extent and impact of discrimination, though perhaps more contentiously he allows for its use as a best proxy in some limited circumstances, identified by a decision tree. For Martin (2005) any attempt to operationalise race/ethnicity for health research fails to meet the important scientific criteria of standardisation and practical utility. It remains the case in our study that many will use the term 'ethnic/family' origins in an uncritical commonsense manner that will reproduce rather than challenge inequalities. Smart (2005) requires that some substance be put upon the judgement criteria proposed by Ellison (2005). These criteria are reliability, accuracy and acceptability. Whilst we do not claim to have solved these dilemmas, this study has attempted to assess an ethnic/family origins screening question for use in a specific health context (assessing risk of carrying genes associated with sickle cell and thalassaemia) in a specific place and time (early twenty-first-century England). The study has attempted to estimate reliability by means of re-interviewing clients; to estimate accuracy by comparing answers to the screening question with laboratory results; and to estimate acceptability by recording the proportion of the antenatal population asked who were prepared to give an answer to a second ethnic question (in addition to an ethnic monitoring question) geared to assessing sickle cell and thalassaemia risk.

In this paper, we have argued that where a more specific purpose is identified, where the categories are elaborated with specific regard to the particular issue at stake, and where the underlying reason for asking the question is fully shared with the participant, then an ethnicity concept may continue to have some utility. Targeted screening for risk of carrying genes associated with sickle cell or thalassaemia may be one such specific instance. However, such screening questions appear to be applied highly selectively by health professionals, and research is also needed to determine what factors are behind this selectivity in asking ethnicity screening questions for sickle cell and thalassaemia in the antenatal context. Meanwhile, it is clear that attempts to minimise the number of real carriers not afforded early antenatal care with regard to screening for sickle cell and thalassaemia depends upon creating sustained educational opportunities for health professionals (primarily midwives) who carry out the screening, on the complex relationship between ethnicity categories and carrier status for sickle cell and thalassaemia, and upon allocation of sufficient time within routine service provision to administer a screening question based on full consultation with the mother about her needs.

Contributors

S.D. was the chief investigator, managed the research, undertook all preparation of ethics applications, contributed to research design, contributed to preparation of tables for the manuscript, took the lead in authoring the paper, and is the guarantor. D.R. contributed to research design and critically appraised the paper. S.H. undertook

the power calculations, all database and data dictionary design, data preparation, data validation, preparation of tables for the manuscript and all statistical analysis. L.A.C. prepared literature for the manuscript and critically appraised documentation at all stages of the research. C.G., A.K., P.M., F.S. and P.S. collected questionnaire and re-interview data; contributed to amendments to the research design; and provided insights to the interpretation of data contained in the manuscript. D.R., A.K., F.S. and P.S. were local lead researchers.

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Competing Interests

No competing interests declared.

Ethical Approval

The EQUANS study was approved by the relevant multi-centre ethics committee and the four local research ethics committees.

Notes

[1] Both mothers and fathers are equally likely to carry genes associated with sickle cell and thalassaemia. The terms of the initial brief given to the research team were to assess the viability of an ethnicity screening question for the mother only. The family origins question later adopted by the NHS Sickle Cell and Thalassaemia Screening Programme addresses a comparable question to both the mother and the father (NHS Sickle Cell and Thalassaemia Screening Programme 2005b).

[2] This was adjusted to 1 in 5 after four months of the study in order to generate sufficient numbers of respondents sufficiently quickly for the study to be statistically viable, in order in turn to produce a report upon which a policy decision depended. Since the first interview was usually at eight weeks gestation and the re-interview was at around 24 weeks gestation, the re-interviews of the last subjects recruited to the study would not have occurred until three months after the end of the main data collection period. This was initially anticipated, but when the study fell behind schedule, this change was implemented so as not to delay results.

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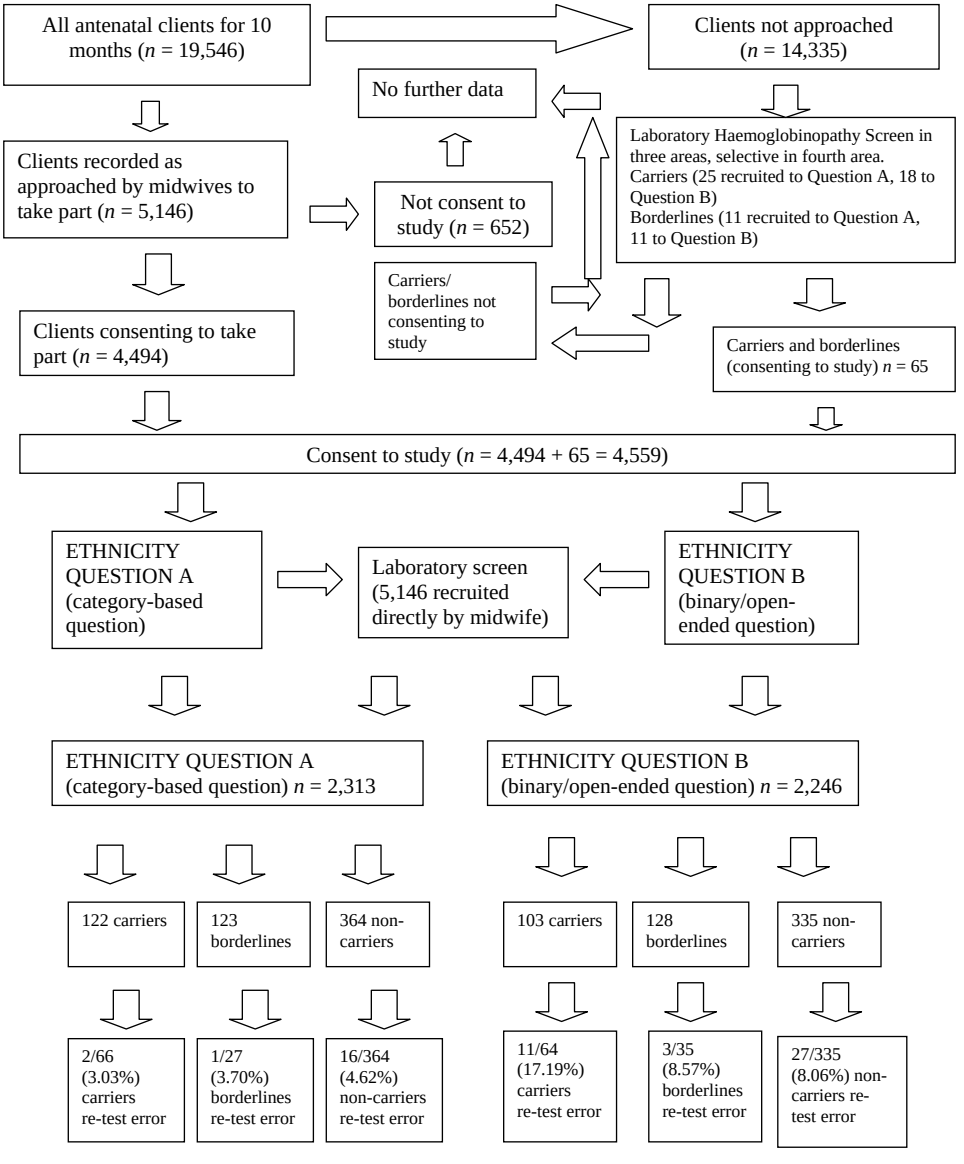

Design of the EQUANS Study