A multidisciplinary approach for improving services in primary care: randomised controlled trial of screening for haemoglobin disorders

Michael Modell, Beatrix Wonke, Elizabeth Anionwu, Maren Khan, Sharon See Tai, Michael Lloyd, Bernadette Modell

Royal Free and University College Medical School, Whittington Hospital, London N19 5NF
Michael Modell, *professor of primary health care*
Maren Khan, *nurse facilitator*
Sharon See Tai, *senior research fellow*
Bernadette Modell, *professor of community genetics*

Whittington Hospital Trust, London N19 5NF
Beatrix Wonke, *consultant haematologist*
Michael Lloyd, *laboratory manager, haematology department*

Mothercare Unit of Clinical Genetics and Fetal Medicine, Institute of Child Health, University College London, London WC1N 1EH
Elizabeth Anionwu, *senior lecturer in community genetic counselling*

Correspondence to: Professor M Modell
m.modell@ucl.ac.uk

BMJ 1998;317:788–91

Abstract

Objective: To investigate the feasibility of improving screening for carriers of haemoglobin disorders in general practice by using a nurse facilitator to work with primary care teams and the relevant haematology laboratories; to identify problems in communication between all those involved in delivering the service, and to implement solutions.
Design: Two year, practice based randomised controlled trial.
Setting: North London area where 29% of residents and 43% of births are in ethnic groups at risk for haemoglobin disorders.
Subjects: 26 of the 93 practices using the services of the area's haematology laboratory agreed to take part and were randomly divided into control and intervention practices.
Main outcome measure: Change in number of requests for screening tests for haemoglobin disorders made by control and intervention practices in baseline and intervention years.
Results: The number of screening tests requested varied from 0-150 in the 93 practices in the baseline year. Study practices tended to have made a moderate number of requests (10-50) during this period. During the intervention year intervention practices made 292 more requests (99% increase) and control practices made 74 fewer requests (23% decrease; $P = 0.001$ for difference in median change). Four practices, three of which were singlehanded, accounted for 75% of the increase. The number of requests from intervention practices, adjusted for baseline requests, was 3.2 times higher than control practices ($P < 0.0001$).
Conclusion: General practitioners and practice nurses are willing to undertake a new genetic screening service (or expand an existing one) if they are persuaded that it benefits the health of a significant proportion of their practice population. They need appropriate tools (for example, information materials for carriers and groups at risk), and the laboratory must be sensitive to their needs. Preconceptional carrier screening and counselling need to be coupled with antenatal screening.

Screening for carriers of thalassaemias and sickle cell disorders (haemoglobin disorders) is established in the United Kingdom,[1] where over 6% of residents and 10% of newborns are of Asian, black, Mediterranean, or mixed parentage.[2] On average, 8% (range 3-15%) of these groups are healthy carriers who can be readily identified by screening for haemoglobinopathy.[3] Carrier couples have a 25% risk in each pregnancy that the child will have a serious haemoglobin disorder, and timely information allows them informed choice among reproductive options, including prenatal diagnosis.

Though the laboratory diagnosis of carriers is relatively simple, screening a population involves many steps and different services.[1] Premarital screening is routine in Cyprus[4] and screening is offered in high schools in Montreal.[5] In Britain most district health authorities have opted for routine antenatal screening. Since maternal blood is often first taken at around 16 weeks of pregnancy, many carrier couples are offered prenatal diagnosis in the second trimester.[6 7]

Diagnosis in the first trimester (by chorionic villus sampling and DNA analysis) is far more acceptable and is feasible for all couples at risk for haemoglobin disorders.[8] When counselled in the first trimester, 85-95% of couples at risk for thalassaemias and 50-80% of referred couples at risk for sickle cell disorders request prenatal diagnosis.[9 10] But only 50% of couples at risk for thalassaemias and 13% at risk for sickle cell disorders actually have a prenatal diagnosis.[6] One reason for the discrepancy is that prenatal diagnosis is often offered in the second trimester.[11] Couples at risk need to be identified before conception, or as soon as they report a pregnancy to their general practitioner.

We tested the hypothesis that it is feasible to promote screening for haemoglobin disorders in primary care by creating a multidisciplinary team and employing a nurse facilitator (MK) to promote the service and improve communication among those involved.

Methods

Study design

Following approval from the relevant ethics committees, a two year randomised controlled trial was carried

out between 1 January 1995 and 31 December 1996 in an area of north London where, according to 1991 census data, 29% of residents and 43% of births are in the relevant ethnic groups.[2] In the baseline year the facilitator was trained, baseline data collected, practices recruited and randomised, and educational materials prepared. In the second year (intervention year) the nurse facilitator trained staff in intervention practices, provided information materials, and reviewed communication between the laboratory and the practices. She strengthened links between primary care teams and the two haemoglobinopathy counsellors.

The main outcome measure was the change in the number of requests for screening tests for haemoglobin disorders made by control and intervention practices in the baseline and intervention years.

Recruitment of practices

Each of 295 general practitioners in 93 practices in 50 wards of the London Boroughs of Islington, Camden, Haringey, Barnet, and Hackney who use the Whittington Hospital's open access haematology service was invited by letter to participate. Those expressing interest were visited by two members of the research team and asked to provide basic information about the practice. Finally 26 of the 93 practices (28%), including 27% of general practitioners, joined the study. Nineteen used only the Whittington Hospital's laboratory and seven also used the Royal Free or University College London Hospitals: these laboratories kindly provided data on screening requests made by the study practices.

The 26 practices were stratified by the proportion of ethnic minority residents in the relevant ward and the number of general practitioners in the practice, as these factors might affect the number of screening requests. They were assigned within each stratum by using computer generated random numbers as 13 intervention and 13 control practices (table 1) in October 1995.

Information materials

Intervention practices were given posters to inform relevant ethnic groups of the existence of the haemoglobin disorders, leaflets explaining why carrier testing is advisable and how it can be obtained, information leaflets for carriers, and a practice reference manual containing background information and copies of carrier information leaflets. Later all general practitioners in the intervention group were sent a laminated card listing ethnic groups to whom screening should be offered to keep in their consulting room.

Educational sessions

Intervention practices were offered three formal 30-60 minute sessions that included a slide presentation on indications for haemoglobinopathy screening, informing carriers, the role of haemoglobinopathy counsellors, and information about local counselling services; viewing the video "From Chance to Choice," which focuses on the role of the primary care team in screening for common recessively inherited diseases and emphasises a multiethnic approach; local statistics for ethnic minority residents and births; a discussion of the reference manual; and, in the final session, a demonstration of the information material generated in the laboratory.

Table 1 Stratification and randomisation of practices

	Intervention group			Control Group		
No of partners	No of practices	<20% ethnic minorities	≥20% ethnic minorities	No practices	<20% ethnic minorities	≥20% ethnic minorities
1-2*	6	1	5	7	2	5
≥3	7	5	2	6	5	1
Total	13	6	7	13	7	6

* Smaller practices (1-2 partners) predominate in areas with a higher proportion of ethnic minorities.

The first sessions were completed at the end of 1995, and the intervention year started in January 1996. The second and third sessions were offered at the middle and end of 1996. Ten practices accepted all three sessions, two accepted two, and one accepted one. The haemoglobinopathy counsellors attended seven of the final sessions. Ten of the 13 control practices accepted the offer of a 30-40 minute training session after the intervention year.

Requests for haemoglobinopathy screening

The Whittington laboratory receives 30 000 haematology requests from general practitioners annually. About 6.5% are for haemoglobinopathy screening, mostly for ethnic minority patients of reproductive age. Screening requests from study, control, and non-participating practices were recorded, using computerised laboratory records. The unit of analysis was number of requests per practice.[12] The Mann Whitney U test was used to compare the change in this number in intervention and control practices during the intervention year. Poisson regression was used to investigate the association between intervention year requests (dependent variable) and baseline requests and a binary variable indicating intervention or control (independent variables),[13] weighted by the square root of the baseline requests to account for the variation[14] between practices.

Results

Self selection of study practices

The number of screening requests in the baseline year varied widely between practices (table 2). Participating practices were representative of local practices in terms of size (38% singlehanded, 42% with two to five partners, 19% with six or more partners) but tended to make a moderate number of requests (10-50 per year) and had a somewhat lower proportion of residents from ethnic minorities (table 3).

Change in number of screening requests

In the intervention year there were 292 more requests in intervention practices and 74 fewer requests in control practices (table 4). This change was significant

Table 2 Haemoglobinopathy screening requests from practices in baseline year

No of requests	No (%) of all practices	No (%) of participating practices
0	14 (15)	0
1-9	37 (40)	7 (27)
10-24	22 (24)	8 (31)
25-49	14 (15)	10 (38)
50-99	4 (4)	1 (4)
100-150	2 (2)	0
Total	93 (100)	26 (100)

Table 3 Proportions (%) of ethnic minority residents, births, and carriers of haemoglobin disorders in wards containing practices using Whittington Hospital laboratory services. Values are means (ranges)

	All wards	Wards with participating practices
Ethnic minority residents	29 (11-55)	23 (12-44)
Ethnic minority births*	43 (19-98)	36 (19-69)
Carriers:		
% of all newborns	6.0 (3.2-14.2)	5.0 (2.5-9.3)
% of ethnic minority newborns	13.7 (9.5-16.5)	13.7 (9.5-16.3)

*Proportion of births exceeds the proportion of residents because of young age distribution and higher birthrate in ethnic minority groups. Prevalence of carriers of haemoglobin disorders is correspondingly higher among pregnant women and newborns.

Table 4 Requests for haemoglobinopathy screening tests in each group of practices[2]

Practices	No of requests in baseline year (1995)	No of requests in intervention year (1996)	Change	Requests in 1996 as % of 1995
Control	328	254	–74	77
Intervention	295	587	292	199

(Mann-Whitney U = 21.5, P = 0.001; median change in the intervention group 8.0, in control group 2.0; 95% confidence interval for median difference 0.5 to 15.0).

There were wide differences between individual intervention practices. Those already making more requests tended to respond more positively (fig 1). The average increase was 50% in practices with < 20% ethnic minority residents and 126% in practices with ≥20% ethnic minority residents. Most practices showed a modest increase, one made fewer requests than before, and four accounted for 75% of the total increase: three of these were singlehanded and the other had four partners (fig 2). Practices that declined some training sessions showed no perceptible change.

The Poisson regression analysis confirmed the positive relation between practices' requests in the study year and at baseline (regression coefficient = 0.025 (SE 0.0009), P = 0.0001), but the association between requests in the study year and being an intervention practice was stronger (1.15 (0.0361), P = 0.0001). The number of requests in the study year for intervention practices (adjusted for baseline requests) was 3.2 times higher (95% confidence interval 2.9 to 3.4) than for control practices.

Communication between laboratory and practices

The Whittington Hospital's haematology laboratory assumed clinical responsibility for distributing information and advice about haemoglobin disorders. The laboratory staff and the research team collaborated to produce a comprehensive, computerised, laboratory based set of information materials. When a carrier is identified the laboratory routinely sends the general practitioners a letter giving the specific diagnosis and an information leaflet, with a request to pass the leaflet on to the patient (information leaflets available at http://www.chime.ucl.ac.uk/APoGI/).

Discussion

This study aimed to encourage carrier screening in general practices in an area with a high prevalence of residents from ethnic minorities. The local haematology laboratory has a tradition of working with local primary care teams, which were requesting on average 6-7 screening tests a year, though 14 practices made no requests and seven made more than 50 a year.

Twenty six practices, almost a third of those using the Whittington hospital laboratory, joined the study. The practices were self selected, with moderate screening activity. No practice that was making no requests collaborated, and six (including three singlehanded practitioners) that were making more than 50 requests a year declined, presumably because they were fully aware of the haemoglobin disorders.

The intervention included the following effective strategies for changing professional behaviour: outreach visits by a nurse facilitator; production and dissemination of educational material for health professionals and patients; and periodic reminders to the general practitioner to screen.[15–17] The approach was successful, with a 99% increase in requests from study practices compared with a 23% fall in requests from control practices. However, the response from general practitioners and practice nurses in the

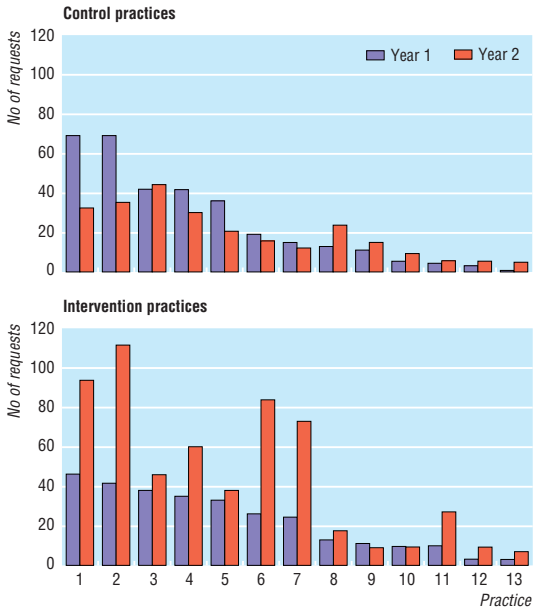

Fig 1 Change in number of requests for screening for haemoglobin disorders in participating practices. Practices are ranked in order of the number of requests in the baseline year

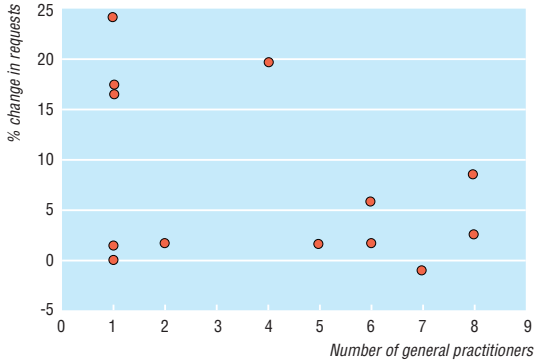

Fig 2 Number of doctors (including GP registrars) in intervention practices and proportion of total increase in requests for haemoglobinopathy screening

intervention group varied widely, reflecting variation before the study began (fig 1).

Factors affecting change in screening

Several factors seemed to affect the change in screening activity.

General practitioner's level of interest—Members of some intervention practices never engaged with the project, nor attended the training sessions: perhaps not all saw it as relevant. Others may have had relatively few ethnic minority patients. Some may have felt uncomfortable with a service that could lead to termination of pregnancy.

Ethnic makeup of the practice population—The increase was greatest in practices in wards with more than 20% ethnic minority residents.

Number of partners—Smaller practices (one or two partners) predominate in areas with a high proportion of ethnic minorities, and 56% of the total change was due to three singlehanded practitioners (3/48 participating general practitioners). This suggests that when a singlehanded practitioner decides to change the effect can be dramatic—a counterbalance to the present focus on the advantages of larger practices.

Feasibility

We conclude that it is feasible to encourage general practitioners and practice nurses to undertake a new genetic screening service (or expand an existing one) if they are persuaded that it is practicable and that it benefits a large proportion of their patients. They need appropriate tools (such as information materials for carriers and groups at risk), and the laboratory must be sensitive to their needs—for example, by reporting results with their interpretation and by issuing prompts such as "sickle cell trait: suggest test partner." Newly identified carriers will need counselling, either within the practice or by a haemoglobinopathy counsellor (there are more than 60 in 40 community based sickle cell and thalassaemia centres[18]). However, the uneven response of the general practitioners shows that additional approaches are needed—for example, a strategy within the maternity service for taking parental blood in early pregnancy—especially in areas with a high proportion of ethnic minority births.

Effects of the study

The study had many effects. The creation of a multidisciplinary team (the authors of this paper) allowed us to examine all facets of delivering the service, including providing information for patients, the demography of practice populations, and communication between hospital and practices. It enabled laboratory staff to respond more precisely to the needs of general practitioners, and it promoted interaction between haemoglobinopathy counsellors and practices. A larger multidisciplinary group has now been formed to review screening for haemoglobin disorders within the local health authority. This is leading to several changes, including regular laboratory based monitoring of the number and origins of screening requests. This will enable us to document the effect of the computerised, laboratory based information materials and monitor the screening activity of individual general practitioners. The effect of the facilitator's brief intervention may only be maintained if practices are prompted at intervals by trained and interested members of the practice team, the laboratory, and haemoglobinopathy counsellors.

Key messages

- Many couples at risk of conceiving a child with a haemoglobin disorder are identified too late in pregnancy for prenatal diagnosis to be acceptable to them
- These couples need to be identified before conception, or as soon as they report a pregnancy to their general practitioner
- Screening for haemoglobin disorders in primary care can be enhanced
- A multidisciplinary approach and a haematology laboratory responsive to the needs of local practitioners are important
- The nurse facilitator was particularly successful in persuading singlehanded practitioners practising in areas with a high prevalence of ethnic minorities to increase their testing for carriers

We thank Denis Menich and Teresa Marlow for help in collecting laboratory data, and Dr John Porter and Dr Atul Mehta from the haematology laboratories at the University College London and the Royal Free Hospitals Trusts. We are particularly indebted to all the general practitioners and practice nurses who took part in the study: this project would, of course, have been impossible without their collaboration.

The video "From Chance to Choice" is available from Mothercare Unit of Clinical Genetics and Fetal Medicine, 30 Guildford St, London WC1N 1EH; it costs £30 (including VAT and postage) payable to ICH Productions plc.

Funding: North Thames Office of the NHS Executive, as part of the National Primary Secondary Care Interface R & D Programme. BM is a Wellcome principal research fellow.

Conflict of interest: None.

1 Department of Health. *Report of a Working Party of the Standing Medical Advisory Committee on Sickle Cell, Thalassaemia and other Haemoglobinopathies.* London: HMSO. 1993.
2 Health Education Authority. *Sickle cell and thalassaemia: achieving health gain—guidance for commissioners and providers.* London: HEA, 1998.
3 British Society For Haematology. Guidelines for haemoglobinopathy screening. *Clin Labor Haematol* 1988;10:87-94.
4 Angastiniotis MA, Kyriakidou S, Hadjiminas M. How thalassaemia was controlled in Cyprus. *World Health Forum* 1986;7:291-7.
5 Ostrowsky JT, Lippman A, Scriver CR. Cost-benefit analysis of a thalassemia disease prevention programme. *Am J Public Health* 1985;75:732-6.
6 Modell B, Petrou M, Layton M, Varnavides L, Slater C, Ward RHT, et al. Audit of prenatal diagnosis for haemoglobin disorders in the United Kingdom: the first 20 years. *BMJ* 1997;315:779-84.
7 Fairweather DVI, Ward RHT, Modell B. Obstetric aspects of midtrimester fetal blood sampling by needling or fetoscopy. *Br J Obstet Gynaecol* 1980;87:87-99.
8 Old JM, Varawalla NY, Weatherall DJ. Rapid detection and prenatal diagnosis of beta-thalassaemia: studies in Indian and Cypriot populations in the UK. *Lancet* 1990;336:834-7.
9 Modell B, Ward RHT, Fairweather DVI. Effect of introducing antenatal diagnosis on the reproductive behaviour of families at risk for thalassaemia major. *BMJ* 1980;281:737.
10 Petrou M, Brugiatelli M, Ward RHT, Modell B. Factors affecting the uptake of prenatal diagnosis for sickle cell disease. *J Med Genet* 1992;29:820-3.
11 Neuenschwander H, Modell B. The process of antenatal sickle cell screening at a north London hospital. *BMJ* 1997;315:784-5.
12 Bland JM, Kelly SM. Statistics notes: trials randomised in clusters. *BMJ* 1997;315:600.
13 Armitage P, Berry G. *Statistical methods in medical research.* 3rd ed. Oxford: Blackwell Scientific, 1994.
14 Donner A, Klar N. Cluster randomization trials in epidemiology: theory and application. *J Stat Plann Inference* 1994;42:37-56.
15 Davis DA, Thomson MA, Oxman AD, Haynes RB. Evidence for the Effectiveness of CME. A Review of 50 Randomized Controlled Trials. *JAMA* 1992;268:1111-7.
16 Davis DA, Thomson MA, Oxman AD, Haynes RB. Changing physician performance: a systematic review of continuing medical education strategies. *JAMA* 1995:274:700-5.
17 Haines A. The science of perpetual change. *Br J Gen Pract* 1996;46:115-9.
18 Anionwu EN. District-based population registers for sickle cell disorders: a role for the haemoglobinopathy clinical nurse specialist? *Child Care Health Devel* 1997;23:431-5.

(Accepted 6 July 1998)