



SCREENING AND COUNSELLING FOR SICKLE CELL DISORDERS AND THALASSAEMIA: THE EXPERIENCE OF PARENTS AND HEALTH PROFESSIONALS

KARL ATKIN,^{1*} WAQAR I. U. AHMAD¹ and ELIZABETH N. ANIONWU²

¹Ethnicity and Social Policy Research Unit, University of Bradford, Bradford, BD7 1DP UK and

²School of Nursing Studies, Wolfson Institute of Health Sciences, Thames Valley University, London, UK

Abstract—Shortfalls in haemoglobinopathy provision result in patients and their carers receiving inadequate support. This paper, by drawing on material from a project evaluating service provision to families caring for a child with a sickle cell disorder or thalassaemia, discusses screening and counselling services. It explores the perspectives of parents, front-line practitioners, managers and health commissioners. Poor quality care, inadequate information and professionals' insensitivity were salient themes in parental accounts. The parents' experience also confirms the problems faced by minority ethnic people in having their welfare needs recognised, more generally. Although our focus is on genetic conditions affecting minority communities in the UK, the issues we address are at the heart of the 'new genetics'. © 1998 Elsevier Science Ltd. All rights reserved

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INTRODUCTION

Research on services for sickle cell disorder (SCD) or thalassaemia major documents a variety of problems; the consequences of which range from the denial of informed choice to avoidable suffering, as well as death (Darr, 1990; Anionwu, 1993; Midence and Elander, 1994; Ahmad and Atkin, 1996a,b). Provision for these conditions also poses complex political and ethical issues (Atkin and Ahmad, 1997). This paper draws on a project evaluating support to families of children with a SCD or thalassaemia[†]; it examines the perspectives of parents and health professionals on diagnosis and screening, within the context of the wider research, policy and ethical debates. Our findings have implications for children with other chronic illnesses and genetic conditions affecting other ethnic groups.

SCDs and thalassaemia major are recessive conditions where individuals who inherit a deleterious gene from both parents develop the disease. In the U.K. the groups most at risk of SCDs are of African-Caribbean and West African origin; thalassaemia is more common among Cypriots, South Asians and Chinese, but also occurs among African-Caribbean and white British people. There are estimated to be between 6000 to 10 000 people

with a SCD and around 600 with thalassaemia in the U.K. There are, of course, many more carriers of these conditions.

SCDs include sickle cell anaemia, haemoglobin SC disease and sickle β -thalassaemia. Those with SCDs are prone to 'sickling' of the red blood cells causing blockages in smaller blood vessels and resulting in 'the painful crisis', as well as anaemia, leg ulcers, stroke and damage to various parts of the body including the spleen, kidneys, the hips, eyes and lungs. Affected children are vulnerable to strokes and life threatening infections, e.g. pneumonia and meningitis. Treatment and care include the prevention of life-threatening infections; pain management and the avoidance of circumstances that cause the red blood cells to 'sickle'. SCDs are variable, unpredictable and, at times, life threatening.

A child born with thalassaemia major is unable to make a sufficient amount of haemoglobin and needs blood transfusions every 4 to 6 weeks, for life. Excretion of excess iron from transfusions requires injections of a drug such as desferrioxamine (desferal), using a battery operated pump, 8 to 12 h a day, 5 to 7 nights a week; death through non-use of the infusion pump is not uncommon. Complications from thalassaemia major, include diabetes, delay or failure to enter puberty and infections, such as hepatitis C, acquired through blood transfusions. In comparison to SCD, the condition is more predictable and stable.

*Corresponding author. Fax: +44-113 2334836.

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SCREENING AND DIAGNOSIS

Diagnosis is a key period for parents of a child with a chronic illness (Eiser, 1990). Early diagnosis enhances the prospects for adequate care, adjustment and coping (Sloper and Turner, 1992; Beresford, 1996). Awaiting a diagnosis is a time of distress and uncertainty (Green and Statham, 1996). The actual diagnosis may evoke feelings of guilt, frustration, anxiety, helplessness and resentment (Atkin, 1992), particularly since parents usually anticipate a 'healthy' child (Richards, 1993).

The genetic nature of some childhood illnesses introduces other important themes. Health professionals experience difficulties in conveying genetic knowledge to lay people; lay understandings of genetic inheritance often differ from those held by professionals (Rapp, 1988; Richards and Green, 1993). Further, in offering pre-natal testing, consultant physicians, for example, are often motivated by the concern to eliminate abnormality rather than encouraging 'informed choice' among parents (Marteau *et al.*, 1992). This tensions between 'informed decision making' and 'prevention' is at the heart of the 'new genetics' and has particular significance for haemoglobinopathies (Atkin and Ahmad, 1997).

Within the context of the broad concerns outlined above, diagnosing haemoglobinopathies raises specific difficulties for parents. As a result of *ad hoc* screening policies, children are often not identified until some time after birth (Davies *et al.*, 1993) and there have been calls for a more systematic screening policy (SMAC Report, 1994; Modell and Anionwu, 1996). Without diagnosis and treatment thalassaemics die before the age of two and up to 30% of deaths in SCD occur before diagnosis (Midence and Elander, 1994)*. In addition, provision of information and emotional support during diagnosis is often poor (Black and Laws, 1986; Darr, 1990), which can prolong uncertainty and potentially undermine a parent's ability to come to terms with the diagnosis (Midence and Elander, 1996). Further, although some localities employ specialist haemoglobinopathy counsellors, counselling and disclosure is often undertaken by health professionals with little formal training in counselling, limited understanding of users' cultural backgrounds and often through poorly trained interpreters or family members. Thus many parents

are denied specialist support and informed choice (Ahmad and Atkin, 1996b). More generally, inadequate haemoglobinopathy provision introduces the issue of racism (Ahmad and Atkin, 1996b). The long term neglect of these conditions by the British National Health Service (NHS) and elsewhere, poor service co-ordination and low priority given to haemoglobinopathies are pointed to by critics and users as reflecting racism (Anionwu, 1993)†.

Here, we use qualitative evidence to examine screening policy and practice, pre-natal diagnosis and informed decision making and communication and information in relation to service provision to affected families.

THE STUDY

This qualitative evaluation of service support to affected families, examined both mothers' and fathers' perspectives on the nature and appropriateness of haemoglobinopathy provision and explored service delivery and co-ordination with front-line practitioners, managers and health commissioners.

We conducted in-depth qualitative interviews with 62 parents of affected children (age range 1–18 yr): 37 had children with thalassaemia and 25 with SCD. This represented 34 mothers, 25 fathers and 3 guardians (an uncle, a brother and a sister-in-law) drawn from health professionals' records in northern England. The parents represented the total population sample of affected families in the seven localities in the study. The coverage across diverse locations and inclusion of the total sample makes the findings particularly robust, by avoiding selection biases. In terms of self-defined ethnicity, 11 families with a child with a SCD were Caribbean; 1 Indian; 1 Algerian; 1 Nigerian and 3 of mixed ethnic origin. For thalassaemia, 15 families were Pakistani; 1 Indian; 2 Bangladeshi and 2 East African Asian.

To gain an understanding of services from provider and commissioner perspectives, we interviewed 51 key service providers, managers and commissioners in the same localities. This sample was generated in one of three ways. First, the families were asked to provide the names of the two most helpful service practitioners; one from health service, the second from an other statutory or voluntary organisation. This allowed an examination of service provision and interagency working. Secondly, key practitioners in each area were asked to put us in touch with senior managers in their local NHS Trust (providers) and health commissioners (health authorities) who had some responsibility for the management of haemoglobinopathy services. Thirdly, as fieldwork progressed, the research team became aware of other practitioners, managers and commissioners whose perspectives it was important to examine. The final sample included consultant physicians, specialist haemoglobinopathy workers,

*Diagnosis of the two conditions is fairly straightforward and involves a simple and cheap blood test. Unlike other genetic conditions it is not necessary to have full family history to detect the condition. It is also possible to test an unborn child as early as twelve weeks in pregnancy.

†Detailed overviews of the practical, social and ethical implications of haemoglobinopathies are available in Davies *et al.* (1993), Midence and Elander (1994), Ahmad and Atkin, 1996b and Atkin and Ahmad, 1997).

genetic counsellors, NHS Trust managers and general medical practitioners (GPs) as well as health commissioners responsible for sickle cell and thalassaemia services. Additional interviews were undertaken with social service managers and representatives from voluntary organisations.

The approach to evaluation adopted by this project required a methodology that could reflect the plurality of social reality and deal with different stakeholders' accounts. We therefore used qualitative methods, based on semi-structured interviews. Such methods allow an examination of complex and contingent situations, behaviours and interactions (Mishler, 1986; Gubrium and Silverman, 1989). A topic guide identified a number of key themes developed from a review of the relevant literature on haemoglobinopathies, chronic illness and ethnicity and welfare, discussions with key informants and advice from an expert advisory committee.

Mothers and fathers (interviewed separately) were offered an interview in the language of their choice and by a researcher of their own sex. The use of gender, ethnicity and language matched interviewers is of help in facilitating access and communication (Ahmad, 1993). Twenty one interviews were in Asian languages (19 in Punjabi, 1 in Bengali and 1 in Urdu). The interviewers were given training in haemoglobinopathies, qualitative interviewing, using the topic guide and translating/interpreting key concepts between English and Asian languages.

All interviews were tape recorded. Those in Asian languages were interpreted into English. Transcribed interviews were organised according to analytical headings. Following accepted conventions of qualitative analysis (Gubrium and Silverman, 1989), information was taken from the transcripts and transferred onto a map or framework, allowing comparison by theme and case. The respondents' accounts were organised by categories and sub-categories, suggested by the topic guides as well as new categories which emerged from analysis of transcripts.

THE PARENTS' AND PROFESSIONALS' ACCOUNTS

We examine how services organise and manage screening for haemoglobinopathies before considering the parents' accounts of diagnosis. A detailed consideration of pre-natal diagnosis and informed decision making is followed by exploration of issues in communication and information, including prac-

tical difficulties of providing genetic information. We then examine explanations of haemoglobinopathies given to parents. The concluding section takes up the main themes and locates them within debates on genetics, ethnicity and citizenship.

Screening for haemoglobinopathies: The response of services

The experience of parents — especially those whose child has a SCD — suggest that diagnosis of haemoglobinopathies was *ad hoc*. The interviews with health care professionals confirmed this. No locality operated a universal screening policy. Selective policies were justified on the grounds of the small at risk populations and limited resources. Few NHS Trusts operated a coherent policy; in most cases, ethnic stereotypes drove screening policy whereby parents were selected on the basis of ethnicity as judged by skin colour or name. A haemoglobinopathy specialist worker* described the policy in her locality:

So the principle in [name of area] is that anybody who's sort of not Northern European origin by appearance or by a name or whatever would be screened before when they book in at pregnancy.

Carriers were often not identified and condition sometimes mis-diagnosed, if the person did not fit these ethnic stereotypes of 'at risk' groups. Mrs Morris†, for example, did not look Asian or African-Caribbean and was thus not regarded as being 'at risk', despite concerns about her daughter:

They kept telling me, all superior like, "Oh no, you are the wrong ethnic background".

Her daughter was eventually diagnosed as having an SCD. Another African-Caribbean mother who knew she carried sickle cell trait married 'a white Englishman'. Unknown to them, the father carried the thalassaemia trait. Despite the child showing typical symptoms of sickle beta-thalassaemia, it took 18 months to test the child as the child did not fit the ethnic stereotype.

Commissioning agencies often added to the confusion either by not knowing about the activities of NHS Trusts or about the condition to steer policy and practice, a problem also noted in other areas of the NHS (Atkin and Lunt, 1996; Flynn *et al.*, 1996). In some cases the commissioner's accounts contradicted those of the front-line practitioners. Many commissioning managers remarked on the general difficulties of finding out the procedures and policies of the provider units and nearly all commissioners claimed they lacked the necessary resources

*Several localities employ specialist health professionals to provide haemoglobinopathy services. There is, however, little consistency over the use of job titles. Sometimes they were known as haemoglobinopathy counsellors, health promotion nurse specialist or project workers. For simplicity, this paper uses the term specialist worker to describe their posts.

†All names used in this paper are pseudonyms.

and specialist knowledge to monitor contract specifications.

Most practitioners, NHS Trust managers and health commissioners, were aware of the problems inherent in screening for haemoglobinopathies and were attempting to formulate more consistent policies but faced problems in doing so. As noted, commissioners lacked knowledge, skills and resources to guide policy and monitor practice. Providers and commissioners were also aware of the difficulties in co-ordinating a screening policy when several different NHS trusts were involved in offering the service. Typically, screening was conducted in acute trusts and counselling was based in community trusts. This division across potential competitors created problems of communication, ownership of information and sometimes fears about divulging sensitive business information. Such problems of inter-agency collaboration are commonly reported in health and social care (SSI, 1997) and haemoglobinopathy provision (SMAC Report, 1994). A paediatric haematologist remarked that the three hospitals in their locality had different policies; there was also no agreed procedure for communicating test results between hospitals. Specialist workers, mostly based in community trusts, could never be confident that the acute NHS trusts were passing all positive results to them for counselling. This raised the possibility that carriers could be identified without appropriate counselling or that counselling was offered by professionals without specialist training (see SMAC Report, 1994; Marteau and Anionwu, 1996).

GPs were particularly criticised; many professionals felt that GPs were not explaining the significance of test results to parents or — as several specialist worker and consultants remarked — not understanding test results themselves. One specialist worker:

It is just that the information the GPs give sometimes is a bit, "Oh, go away, it is all right, there is nothing to worry about" and people want more than that... I have had instances where I have sent a letter off to somebody, saying I am going to come and see you to discuss the results and they have said, "Oh there is no need to come, doctor said, ...there was nothing to worry about".

Other practitioners and commissioners complained that the fragmentation of ante-natal services and the increasing importance of primary health care in pregnancy hindered communication, especially for the first pregnancy. A paediatrician remarked:

It is not helped by the changes in childbirth which had diversified ante-natal care out of the relatively tightly controlled consultant based care into more diverse community

based care. And realistically, I do not think we would score many points for a first pregnancy effort in ante-natal diagnosis. There are too many people involved.

Several felt that GPs referred patients to hospital services too late for it to be of value for screening and informed decisions by parents. Research evidence supports this view, indicating insufficient monitoring of minority ethnic pregnant women in primary health care (Parson *et al.*, 1993); late referrals to hospital ante-natal care (Jain, 1985) and poor quality provision (Clarke and Clayton, 1983). Evidence also suggests that GPs do not fully understand the value and purpose of genetic testing (Royal College of Physicians, 1989).

Another problem identified by professionals who specialise in haemoglobinopathies as well as health commissioners was the lack of understanding among health professionals generally about the relevance of screening. This was of particular significance given the discretionary nature of screening policy. Parents also felt that professionals were not knowledgeable about haemoglobinopathies (see Midence and Elander, 1994; Green and Murton, 1996). Many felt that a successful policy also required greater understanding of haemoglobinopathies among the affected communities. Many health professionals thought that not all prospective parents recognise the value of screening or of carrier status or understand the mechanisms of genetic inheritance. The need for community education programmes to increase awareness of haemoglobinopathies was emphasised by some. The literature confirms Asian and African-Caribbean population to have a limited understanding of haemoglobinopathies (Anionwu, 1993; Dyson, 1997), findings consistent with lay knowledge of genetically inherited conditions more generally (Green, 1992).

The process of diagnosis: The parents' accounts

Diagnosis of thalassaemia offered few problems and is not discussed in this section. In contrast, confirming the general literature on haemoglobinopathies (Davies *et al.*, 1993) and other genetic conditions (Green and Murton, 1996), diagnosing SCD was problematic and preceded by a period of 'mysterious' symptoms (e.g. mobility problems, constant infections, stomach pains, painful joints, swollen stomach, hands and feet). In retrospect most could be recognised as typical SCD symptoms. Most children were diagnosed as having SCD between the ages of 2 and 3 yr: however, 3 were aged 10 or over when diagnosed.

Several parents experienced lengthy period of misdiagnosis; having to cope with the stress of not knowing what was wrong with their child (see Sloper and Turner, 1992). For three, the process of (mis)diagnosis went on for several years. Legitimate concerns were often dismissed and parents had to

challenge health professionals' opinions to be taken seriously. Since mothers were more often in contact with services they were more likely than fathers to challenge medical professionals, especially. Fathers were also more likely to accept that the doctors, in the words of one father, "knew what they were doing". Mothers, usually having greater knowledge of the child's condition than fathers, were more suspicious of the medical explanations as they often did not correspond with their own experience.

The struggle to be taken seriously was distressing for many mothers who felt undermined by the diagnostic process. Many concerned mothers were dismissed as 'neurotic' and 'over-protective'; some were offered counselling for their 'morbid' obsession with the child's health. Gender as well as ethnic stereotypes, were very much part of the medical responses to parental concerns; this is consistent with other research findings (McIntyre and Oldham, 1977; Twigg and Atkin, 1994); the need to acknowledge and respond to parental concerns about their children in emphasised in various recent publications (e.g. Beresford, 1996).

Dismissal of mothers' concerns is evident in several examples. One mother, for example, was 'made to feel' 'hysterical and uninformed', 'obsessed by SCD', when she insisted on a test after her son suffered several unexplained bouts of illness. Mrs Morris felt her daughter was prone to infections and seemed small for her age. For 4 yr, she was reassured there was nothing seriously wrong. During this process, her daughter's 'failure to thrive' was attributed to 'poor parenting skills'; she was also offered counselling for her 'morbid obsession' with her daughter's health. Eventually, her persistence paid off and following a test, a diagnosis of SC disease was confirmed. As noted, many mothers felt their treatment to be racist and sexist.

More generally, parents of a child with a SCD usually regarded diagnosis as something that had to be fought for (see also Green and Murton, 1996). Further, the mothers' experience — confirming the work of Hill (1994) — meant they often distrusted their future contact with health professionals. Mothers reasoned that since health professionals demonstrated incompetence during diagnosis, they could not be trusted to offer appropriate future care. Further, having a confirmed *diagnosis*, did not end parental problems. Several commented that health professionals did not seem to know much about SCD and thus could not be trusted to give good care; a situation some attributed to SCD being a 'black condition'. This meant that they had to acquire their own information and again challenge health professionals to ensure their child received adequate care. For example, Mrs Johns waited 3 yr for her son's diagnosis of sickle-beta thalassaemia and was then, wrongly, told by her consultant that the condition does not have serious consequences. The mother 'educated' the consultant

by showing medical journal articles to ensure adequate treatment.

The struggle of obtaining diagnosis made many parents assertive and pro-active in dealing with professionals (see also Hunter *et al.*, 1989). As has been reported for other genetic conditions (Green and Murton, 1996), interaction with professionals (Twigg and Atkin, 1994) and patient-professional communication, parents with better education and who are articulate were more able to be assertive. Several parents also commented on their ethnicity, feeling that white health professionals were used to 'fobbing off black people and treating them as though they were 'thick''. Another mother remarked:

They think you are incapable of understanding, you know... that is the irritating thing about it.

Such views about racial marginalisation were also shared by parents of children with thalassaemia and are supported by research literature (Ahmad *et al.*, 1991; Bowler, 1993; Ahmad and Atkin, 1996a,b).

For their part, specialist practitioners, managers and health commissioners were aware that some front-line practitioners had limited or outdated knowledge of haemoglobinopathies and that on occasions this resulted in an unacceptably poor quality of care. Many agreed that basic presenting symptoms of SCD were sometimes missed by doctors and that care after diagnosis could be improved. Consultants, who saw few parents, found it especially difficult to keep up to date with current developments in diagnosis and care. The policy guidance on haemoglobinopathy provision supports this view (Davies *et al.*, 1993; SMAC Report, 1994). One haematologist working in a general hospital, with few minority ethnic patients, remarked:

I am not surprised we miss things really. You see we do not see that many people with sickle cell or thalassaemia. And maybe its not the first thing on your mind when you see a sick child for the first time.

Non-specialist staff and junior practitioners were thought by specialists and parents to have particularly limited knowledge of haemoglobinopathies.

Pre-natal diagnosis and informed decision making

Pre-natal diagnosis was rare among parents of children with a SCD. This, however, did not necessarily reflect a reluctance on the part of these parents to consider such a test, but was more a consequence of inadequate screening policies.

On the other hand, professionals' assumptions that Asian parents find termination unacceptable

may have led to pre-natal tests being denied to some Asian parents (Atkin and Ahmad, 1997). However, practitioners' suspicions about certain parents having affected pregnancies may explain why, in many cases, thalassaemic children were diagnosed very swiftly. Occasional problems, however, did occur; similar to those described by mothers whose child had SCD. One mother described how her concerns were dismissed by a GP and health visitor for 3 months. It was only when the child became seriously ill, with jaundice and diarrhoea, that the GP took her concerns seriously and referred to hospital. Nonetheless, for thalassaemia, health professionals often suspected a couple were at risk of giving birth to a child with thalassaemia and monitored the situation without always informing parents. Parents were generally satisfied with the speed and process of the diagnosis; although not kept informed about the tests was a frequent complaint of parents. One mother said that it was bad enough knowing your child was ill, without having to keep asking the hospital staff what they were doing — this concern is also noted in research on children with a chronic illness (Beresford, 1996) and with genetic condition (Green and Murton, 1996).

Generally, professionals' assumptions about South Asian patients' refusal to terminate affected pregnancies led to the denial of choice to many Asian parents, thus reflecting the role of cultural myths in undermining accepted good practice (SMAC Report, 1994; Ahmad and Atkin, 1996a). South Asian people's responses to termination are as variable and complex as any other section of the population (Green, 1992). We found that termination was acceptable to some families but not to others and further, that some parents may make different decisions during different pregnancies: something also noted by Darr (1990). Indeed the most common reason for turning down pre-natal diagnosis was the timing of the test, often during the second trimester. Late testing was unacceptable to most of the South Asian families interviewed in the study and confirms other research findings (Modell and Anionwu, 1996). To this extent, South Asian families are perhaps no different from the general population (Richards, 1993). Yet many practitioners falsely attributed this reluctance to accept pre-natal diagnosis to religious objections to termination. This illustrates the limitations of applying cultural stereotypes when offering health and social care and emphasises the need to ensure early booking for ante-natal care (Clarke and Clayton, 1983). Several families' views on termination also changed over time. A third of the families who have a child with thalassaemia, would consider prenatal diagnosis and the possibility of termination in future pregnancies. In some cases, this was despite opposition to termination during the first pregnancy.

For many South Asian families the wider family was an important reference point in decisions about pre-natal diagnosis and the possible termination. This was rarely the case among the few African-Caribbean parents offered the choice of pre-natal diagnosis; they usually discussed the situation among themselves without involving the extended family. South Asian mothers and fathers either talked their options over with their own parents or siblings or decided that their families would be so against pre-natal diagnosis and the possibility of termination that it was not worth pursuing the option. There was no difference between those parents who shared a household with their family and those who did not. For example, Kausar Rasool had considered a termination but said that her parents and in-laws influenced her eventual decision not to. Hajira Malik was initially prepared to go through a termination, but refused 'at the last minute'. Her initial decision to terminate had caused tensions among her own and her husband's family. Pressure from other family members was the eventual reason she did not go ahead with the termination:

I remember them all saying "God gave her this child"... The reason why I did not have it done is because the day before [the proposed termination] my father phoned me up and he said to me "if you go ahead with the termination we will never talk to you again", so I "phoned up the next morning and cancelled".

However, not all families were opposed to termination or were able to influence decisions. In one case a mother, with a child with thalassaemia whose earlier termination had led to family conflict, did not inform her husband, in-laws or parents about her decision to terminate:

My husband, he was totally dead against termination... I did not tell him. I did not tell him, no way.

The family accepted the loss of the baby as a miscarriage. Consequently, the mother went through, what was for her a traumatic experience, without the support of her family.

The few families at risk of giving birth to a child with either a SCD or thalassaemia, who had pursued pre-natal diagnosis felt health professionals expected them to abort an affected child. Several were reminded about the seriousness of the condition by health professionals — with hindsight some thought professionals' exaggerated the seriousness of the condition to influence parental decisions — and informed that termination was the best option for the child. Gulab Mohammad remarked:

Because at the time we took it really serious, you see, the doctor scared us, I mean he says that he [the baby] can have all blood transfusions and everything and we thought that it was not going to be a really good life for him.

In some cases parents were actively discouraged by health professionals from going ahead with a test if they had no intention of terminating an affected child. Several families, for instance, were told about the 'considerable' risks of spontaneous abortion following pre-natal diagnosis* and the cost of pre-natal diagnosis. Shahzad Fazal, for instance, related how their consultant emphasised the cost of the test, saying it would be a waste of NHS money to agree to pre-natal diagnosis if they were not going to abort an affected child. One African-Caribbean family had aborted an affected child, following a pre-natal diagnosis: a decision they now regretted. They felt the consultant forced them in to a termination. The mother explained:

The doctor or consultant, whoever it was, he was more or less leading us towards a termination by saying "why have a child like that when you can, sort of you know, pick and chose and have a normal healthy child".

Some time after the abortion, the father — now better informed, after reading books on SCD — came to the conclusion that the consultant offered an extremely biased account of SCD. He discovered that the condition was more variable than they were led to believe:

Well I did not agree with the first abortion. I regretted that, because at the time we did not really get enough information about sickle cell disease and we was under the impression that sickle cell was a life or death sort of disease and the advice was given to have the abortion.

Several consultant physicians confirmed these parents' accounts and were often motivated by the concern to eliminate abnormality rather than encouraging informed choice (see Atkin and Ahmad, 1997, for a discussion of these issues). This approach confirms research evidence on other genetic conditions: for example, 33% of obstetricians get the patient to agree to the termination of an affected pregnancy before proceeding with amniocentesis of chorionic villus sampling (Green, 1995). Several doctors, drawing on their professional socialisation, emphasised their role in preventing illness and could not understand why parents would pursue pre-natal diagnosis if they were not going to

abort an affected child. Their concern was especially strong in the case of thalassaemia. A consultant explained:

Well certainly with sickles I would screen both parents and if both parents are carriers then I would explain the significance of having a child with sickle cell. Explain to them and leave it to them to decide whether they are going to plan a family or not. As far as thalassaemia is concerned, I am probably more directive in that I do consider that the condition has more morbidity and so I do tend to press for pre-natal diagnosis if they are going to have a family. I would then suggest termination.

Another consultant adopted a similar approach and encouraged parents at risk of a child with thalassaemia to undergo pre-natal diagnosis:

I suppose I do not outline all the options as being neutral, because I think maybe the families don't understand how bad it is going to be to have thalassaemia. I do go out of the way to make the option of ante-natal diagnosis and try to stop them, you know, 'Keep our fingers crossed' is likely to be the response and I like to encourage them to think more seriously about the other options. But I do not believe there is any such thing as value free counselling, it is not about a neutral choice and I feel I need to lean on their background prejudices by encouraging them towards... one view rather than another. So I have to admit that I do advocate termination, I suppose.

At a social level, these consultants' assumptions were beginning to extend medicine's claims of competence to areas of personal and social life (Zola, 1975; Atkin and Ahmad, 1997). This was especially evident as some consultants were explicitly advocating particular types of 'appropriate behaviour' on the part of patients who are seeking or should 'be' seeking genetic services.

Parents, on the other hand, usually wanted to use pre-natal diagnosis to seek assurance of the 'normality' of the baby or prepare themselves for possible birth of an affected child. To this extent parents at risk of sickle cell and thalassaemia seem to share similar views to parents at risk of other genetic disorders (Richards, 1993). Nusrat Nabi, for example, explained why she had her three children tested:

I suppose in a way it is better to find out real soon, rather than awaiting a few months going through all that worry.

For parents, termination of pregnancy in case of an affected baby was never the main or only, reason for requesting pre-natal testing. This difference in the views of health professionals and parents is noted in the more general literature on genetic con-

*There is a one in hundred risk of spontaneous abortion from pre-natal testing.

ditions (Rapp, 1988) and reflects a 'mismatch' between professional logic and lay views (Marteau *et al.*, 1992).

The differences in parents' and consultants' perspectives on termination were reflected in the more general tension at the heart of the 'new genetics': the tension between informed decision making and prevention (Clarke, 1991). As we have seen, several consultants have no difficulties in advocating termination. This view, however, was not common among all consultants or other health professionals. A paediatrician, for example, emphasised the importance of allowing parents to make up their own minds:

If [ante-natal diagnosis] is pursued and the child is affected, most families opt to keep the child. I will support their decisions. It is entirely up to the parents...It is got to be their decision.

Many health professionals agreed that counselling should be non-directive, enabling parents to make an informed decision. A specialist worker, for example described her role:

We see ourselves as you know, advocates, you know ensuring that people are able to make informed choices about their future. We are not here to just ensure that there is no thalassaemia major babies born ever again or sickle cell babies. It is not about that, our interests lie with the families and ensuring that they are given the appropriate, adequate information to be able to make informed choices.

These health professionals were especially worried about other health workers advocating termination and could provide examples — confirming the accounts of parents — where the principles of informed decision making were not followed. The ability of health professionals, not formally trained in offering genetic counselling, to offer advice to parents compounded the problem (also see above).

Health commissioners generally claimed to support the principles of informed decision making. Several, however, who advocated informed decision making, maintained that a screening and counselling service was cost-effective because it reduces the incidence of haemoglobinopathies*. This tension between informed decision making and prevention is also apparent in Department of Health recommendations (SMAC Report, 1994). A commis-

sioner manager, for example, described the agency's approach to counselling:

Empowering them [parents] to make their own decisions. *And to prevent* by giving people an informed choice. (our emphasis)

This tension was especially apparent among health commissioners attempting to establish a screening service. These commissioning managers and public health consultants often had to convince the whole commissioning agency about the value of the service. Emphasising the cost-effective nature of provision provided an opportunity to do this, particularly when there were so many other priorities for the health commissioner to consider. One public health consultant felt that the idea of termination was emphasised to attract the commissioning agency:

The way it was pushed was in terms of potential health in the future... Certainly from the point of view of thalassaemia, to prevent a case, in other words terminate a case than to, for a baby to be born and to need care throughout [is more cost-effective].

Communication and information

Appropriate information can facilitate successful coping, as parents make sense of the condition and utilise or obtain resources (Baldwin and Carlisle, 1994). People with SCD or thalassaemia, however, often complain about a lack of information and express doubts about the ability or willingness of health professionals to provide such information (Darr, 1990; Midence and Elander, 1994). Most parents in this study received some information once their child was diagnosed. (Two parents claimed to have received no information whatsoever.) The nature and quality of this information was, however, variable. Those in contact with a specialist worker usually spoke more highly of their experience, than those who were informed about the condition by a consultant. Parents in contact with specialist workers, for example, felt the condition was well explained and found workers approachable. On-going contact with workers was particularly valued, as explained by Mrs Leigh, whose child had SCD:

I mean there is a lot to take in at the beginning. An awful lot, know what I mean? What do you do? You want to be told about a crisis when it is happening, not six months before. It is only then you understand... I mean it is useful to know about crisis. I am not saying it is not. But you have many more questions when you are just been through one.

*To help commissioners in this calculation, there are costing models demonstrating the chapter costs of screening compared in the costs of providing life-long care for a child affected with a haemoglobinopathy (see Ostrowsky *et al.*, 1985).

A specific advantage mentioned by South Asian parents was a common linguistic and cultural background; we return to this later.

By contrast, most parents expressed dissatisfaction with their initial contact with consultants, although some consultants, usually those with a specialist interest in haemoglobinopathies, were praised for their sensitivity and competence. Common complaints included confusing, inadequate or rushed explanations, an inability to ask questions and a failure on part of consultants to volunteer information. Mrs Hardin's response was typical:

If it was up to them [consultants], you would know nothing. You have to ask the questions. They are not going to tell you anything without you asking. Not if they can help it.

A one-off information session was therefore never adequate (see Sloper and Turner, 1992 and Green and Murton, 1996 for discussion of Duchenne muscular dystrophy and childhood disability, respectively who also suggest that the manner of the person giving the information and the ease with which parents felt they could ask questions were key determinants of satisfaction).

Problems in obtaining information were compounded for those who could not speak or understand English. Language support was generally poor (see also Butt, 1994; Walker and Ahmad, 1994) and information was often only available in English. Parents were sometimes offered an interpreting service inappropriate to their language needs. Nurun-Nissa Hussain was a Bengali speaker in an area where there was only Punjabi language support available. This meant she could not understand her daughter's treatment. Interestingly, her refusal to accept the Punjabi interpreter provided by the hospital, was seen by the consultant, as sign of her being difficult. Sometimes family members were used as interpreters and, although acceptable to some parents, others objected to the practice. In some cases children as young as eight years were expected to interpret complex medical information. Several mothers also pointed to the problems their husband faced in simultaneously translating distressing information and coming to terms with it themselves. Fathers confirmed these problems and had particular difficulties in deciding how much they should tell their non-English speaking wives: often they wanted to 'protect them' from information deemed upsetting. However, this often left mothers without important information about thalassaemia; information important for understanding, coping and caring. This is especially important because these mothers usually took on the main responsibility of care, yet were dependent on another gate-

keeper for information about the condition. This contrasted with the situation of mothers whose child had SCD; they had direct contact with practitioners. Consequently, many Asian mothers, were left without a 'voice' and this is why specialist workers who shared the linguistic and cultural background of these women were especially valued.

Difficulties still occurred when interpreters were used (Chamba *et al.*, 1998). Most interpreters, for example, had little specialist knowledge about thalassaemia and faced difficulties in interpreting clinical information and procedures, sometimes with unfortunate consequences. Robina Javed remarked that poor translation meant they did not understand that thalassaemia was a life long condition. Nor did the Javed family realise that thalassaemia was inherited until after the birth of their third child with the condition. Previous interpreters had emphasised that the child was born with the condition, not that it was transmitted through the parents. Other parents felt the process of interpretation inhibited discussion, making it difficult to ask questions. To this extent, the interaction with the health professional seemed artificial. This is another reason parents valued specialist workers who shared their cultural and linguistic background.

Many practitioners shared these concerns. For example, interpreters were often not available or else difficult to organise; something noted by several consultants. For these reasons, many health professionals preferred to use family members. Even when interpreting services were available, practitioners still identified problems. Several practitioners remarked that there were often differences in the language or dialect spoken by the interpreter and that spoken by the patient. Others questioned interpreters' skills in interpreting genetic information as well as their own competence in working with interpreters (see also Chamba *et al.*, 1998).

Several health professionals recognised, however, that barriers to communications were more than language specific. A paediatric haematologist, for instance, described the general problems associated with communication:

I think once you get into things like counselling, say particularly the Asian community where, let us face it, their whole culture and their cultural background is very different from ours.

The issues raised by communication led several practitioners to comment on the importance of recognising cultural differences when providing information, as noted by a paediatric out-reach nurse:

There have been quite a few occasions when I have felt I have needed to be the advocate for the patients and point

out that these families do come from a very different background and culture and just because it's different does not mean it is wrong.

Except for specialist workers, most professionals felt ill-equipped to respond to cultural differences. The workers' understanding of cultural perspectives and language skills helped them offer more tailored information and culturally appropriate support. Mothers also appreciated being able to talk to someone who shared their own gender: especially important considering these mothers were often marginalised during the process of service delivery. The obvious value of specialist workers could not, however, mask a more general problem facing all practitioners: the difference between professional and lay perspectives on genetic inheritance.

The practical difficulties of providing information

Many practitioners commented on problems in explaining genetic information to parents who often knew little about genetics or had different conceptions of inheritance or risk (see Rapp, 1988). These difficulties, as we have seen, were compounded by language and cultural differences. One specialist worker, for example, described the problem of translating basic concepts:

It is like sort of, it is when you are sort of trying to, especially with the screening, for example. Even with thal' majors really. It is like finding the word. Which there is not a word which really actually exists. I use, I sort of, I sort of use things that they use in daily living in order to sort of convey that information.

Nor did having 'words in common' resolve the problem, as noted by a consultant in relation to the notion of statistical risk:

I think we have as much problem with the understanding barrier where we have words in common but not necessarily concepts in common or beliefs in common that we are telling the risks one in four and that means that the fact that they have two infected children does not mean the next one and the next six are going to be all right. Just like people are buying lottery tickets they are understanding of chance is relatively unsophisticated.

Several parents' understanding of risk fitted this model and underlines the disparity in lay and professional discourses of genetics (Rapp, 1988; Atkin and Ahmad, 1997).

Parents understanding of risk further illustrates the differences between lay and professional perspectives, as well as between parents. Some regarded a one in four chance of giving birth to a child with a haemoglobinopathy as an acceptable

risk; others felt the risk was high and would opt for pre-natal diagnosis. In many cases, a parents' sense of risk was also informed by an idea of 'fairness'. Mrs Evans, for instance, believed there was little risk of giving birth to another child with a SCD, because it would be 'unfair'. Other parents held similar views. More generally, a parents' understanding of inheritance, genetics and risk did not seem related to their ethnicity or gender. Further, most parents' understandings, although complex, did not mirror formal discourse on genetics.

Thalassaemia and consanguineous marriage

The process of communication, however, could be further corrupted by health professionals over-emphasis on possible 'causes' of thalassaemia. Few families could provide an explanation of thalassaemia or its consequences. A third of parents, for instance, remained unaware of the genetic consequences of thalassaemia. However, despite the generally poor understanding, most parents associated thalassaemia with consanguineous marriage. Two-thirds said they were explicitly told this by professionals; a claim confirmed by professionals. A paediatrician explained:

The majority of them are of Mirpuri Pakistani origin and of those the great majority of those are consanguineous. So they are quite a high risk population despite their small size, that is why we get so many children with thalassaemia. We try to talk them out of it [marrying cousins] but they would not listen.

Other health professionals, however, were aware of the limitations of this approach. One specialist worker said:

It is just the way in some areas where it creates, I suspect a lot of misguided guilt and distrust because people were blamed, I think, probably unnecessarily and inappropriately.

More generally, the association between thalassaemia and consanguineous marriage is perhaps unfortunate, because it was one of the few 'facts' about thalassaemia that families could remember. Such information caused considerable guilt among parents, as they felt responsible for their child's condition. This guilt was largely absent from the accounts of parents of a child with a SCD. As a consequence of this guilt, South Asian parents also found it difficult to come to terms with the diagnosis. Hajira Malik, for example, remarked that such guilt never really disappeared. The difference in what parents were told by professionals about causation is interesting considering that the nature of transmission of both SCD and thalassaemia is iden-

tical. More generally, the emphasis on consanguineous marriage among health professionals, is seen by some commentators as an example of the implicit racism in medical discourse, in which the health problems of people from ethnic minorities become located in their presumed cultural and biological pathology (Darr, 1990; Ahmad, 1996).

CONCLUSION

The findings substantiate the problems described in haemoglobinopathies literature (Midence and Elander, 1994) and research on other genetic disorders (Green and Murton, 1996). Themes such as poor quality care, inadequate information, insensitivity to the parents' worries and failure to meet their needs re-occur throughout the analysis. The parents' experience also confirms the substantial literature describing the difficulties minority ethnic people face in having their health care needs recognised and catered for (Ahmad and Atkin, 1996a). Equally, parents' high regard for specialist haemoglobinopathy workers reminds us that services can be sensitive and empowering.

Parents of children with SCD, had particular difficulties in being taken seriously. In this respect, diagnosing sickle disorders seem no different from other childhood chronic illnesses (Beresford, 1996) or genetic conditions (Green and Murton, 1996). In a study examining the diagnosis of Duchenne muscular dystrophy, for example, parents found it difficult to convince professionals that there was something wrong (Green and Murton, 1996). Gender, lack of assertiveness and education background were found to be important variables in the process of diagnosis. General work exploring welfare provision (Twigg and Atkin, 1994) support this. Many mothers in this study felt they suffered both sexist and racist marginalisation.

Many parents felt that haemoglobinopathies would have a higher priority if they were not 'black conditions'. Ethnic and racial stereotypes were important in driving screening and counselling policy and interaction with parents. There was also evidence that patient choice was limited by the often racialised and stereotypical perspectives of service professionals or their inability to provide linguistically and culturally sensitive services. The general literature supports these problems, suggesting that identification of genetic conditions cannot be divorced from the racist service provision to minority groups or from historical legacy of eugenics (Bradby, 1996; Atkin and Ahmad, 1997). Many parents felt that racism explained the low priority and poor co-ordination of haemoglobinopathy services and professionals' negative attitudes.

The findings reported here also have more general relevance. The increasing interest in the genetic basis of disease and the development of provision to identify such conditions means that many of the

issues raised by this paper are at the heart of the 'new genetics' (Richards, 1993) and are relevant to other ethnic groups. To this extent, this paper illustrates the general complexities and difficulties of screening and diagnosing recessive conditions, confirming many of the problems noted in the general literature (Clarke, 1991). Evidence from this study, for instance, supports research on other recessive conditions, suggesting that screening and counselling, whether pre-conceptual, pre-natal or ante-natal is often *ad hoc* (Modell and Anionwu, 1996). Practical problems included health professionals' poor knowledge of the value and purpose of genetic screening; late referral of pregnant women for pre-natal diagnosis; the lack of coherent screening and counselling policy; poor interagency collaboration; and a primary health care service ill-prepared to provide screening and counselling.

Besides these practical considerations, this paper raises important philosophical issues facing screening and counselling provision. The first concerns the tension between 'prevention' and 'informed decision making'. The parents' experience shows denial of informed choice and services biased in favour of 'prevention'. The perspectives of many professionals raise questions about whether value-free counselling based on the principles of informed choice is possible (see Clarke, 1991). It seems impossible to avoid normative assumptions about the undesirability of certain conditions and of giving birth to children with these conditions (Stacey, 1996). When screening is offered, senior professionals steer parents towards termination of affected pregnancies (see Green and Statham, 1996; Atkin and Ahmad, 1997). Although many professionals, especially specialist workers trained in counselling, felt that counselling should be non-directive, several consultants, advocated termination of affected pregnancies. The Royal College of Physicians (1989) implicitly supports these assumptions by regarding the birth of a child with a genetic condition as a medical failure. Health commissioners, although supporting the idea of informed decision making confused the debate by emphasising the cost-effectiveness of screening on the assumption that parents will opt for 'prevention' of affected births.

This mismatch between professional and lay logic about genetics we report, is confirmed in the general literature which shows differential discourses of genetic knowledge, especially inheritance and risk (Rapp, 1988; Richards, 1993; Richards and Green, 1993). The 'mismatch' is also evident in other aspects of the communication process. The difficulty of communicating genetic information were compounded by language and cultural differences. Individuals do not make simple choices in light of relevant information; a variety of personal and social factors influence decisions, as we have noted (see also Michie and Marteau, 1996). Those offered

tests, for example, may know little about the disorder they are invited to avoid, distrust medical wisdom or have other reasons to ignore medical advice. People, for example, have difficulty coming to terms with the idea that a healthy individual (a carrier) can pass on a genetic condition (Rapp, 1988). In the case of cystic fibrosis, for example, carriers may not accept it as a genetic condition but still hold it runs in families (Richards, 1993). As we have seen ideas of risk are also not uniform; statistical risk is not always easily accommodated in lay knowledge and concepts such as 'fairness' also come into play.

To conclude, policy and practice needs to ensure the difficulties faced by families with children with a haemoglobinopathy are addressed. These include the need to respect the concerns of parents as 'experts' on their own children (Beresford, 1996); a coherent and comprehensive screening and counselling policy; improved training of front line practitioners and better provision of information to parents. Similar issues emerge in the diagnosis of other childhood chronic illnesses (Beresford, 1996) as well as other genetic conditions (Green and Murton, 1996). Further, service priorities, cannot be isolated from the general debates about providing genetic screening services for the general population. The tension between informed choice and prevention is central to this debate, as is the difference between lay and professional perceptions of genetic inheritance and risk. Nor can these priorities be divorced from debates about service provision to minority ethnic communities, especially since racialised stereotypes and the inability to meet the linguistic and cultural needs of affected groups characterise haemoglobinopathy services.

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