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Social adjustment in three cultures: data from families affected by chronic blood disorders. A sibling study

C. CLEMENTE,¶ J. TSIANTIS,† I. KOLVIN,* G. BA,‡ S. CHRISTOGIORGOS,† C.LEE§, B. TAYLOR¶ and R. MILLER§

**Tavistock Centre, Child and Family Department, London, UK; ‡Psychiatric Department, IRCCS Ospedale Maggiore, Milan University, Milan, Italy; §Haemophilia Centre, ¶Community Child Health, Royal Free Hospital School of Medicine and University College, London, UK; and †Aghia Sophia' Children's Hospital, Athens, Greece*

Summary. This study explores the rate of psychosocial dysfunction in affected and unaffected children from families with haemophilia or β-thalassaemia, as part of a cross-sectional, multicentre study into the resilience of 115 families with blood disorders. Sociodemographic and developmental data were collected from the parents using a standardized and semi-structured interview format, and medical data were obtained from the clinician. The children's social functioning over the year prior to the assessment was assessed with The Social Adjustment Scale adapted for school-aged children. Children with β-thalassaemia showed significantly higher rates of social dysfunction than their unaffected siblings or children with haemophilia and their siblings. Older children showed significantly higher social dysfunction at school. The high rate of social dysfunction in children with β-thalassaemia compared with unaffected siblings is likely to have a basis in the negative experiences associated with their medical problems. In contrast, the therapeutic advances in haemophilia allows boys to lead an almost normal life. Overall, the rates of social dysfunction in families with both these disorders proved commoner than reported in population surveys, but with the unavailability of local population controls, caution needs to be exercised in the interpretation of this finding.

Keywords: haemophilia, siblings, social adjustment, β-thalassaemia

Introduction

Haemophilia and β-thalassaemia are both inherited disorders. Those suffering from haemophilia may have bleeding episodes following trauma or even spontaneously, depending on the severity of the condition. The introduction of treatment with factor replacement has improved the quality of life and lengthened life span. Before the introduction of effective treatment, there were commonly long periods of convalescence following bleeding episodes. Inevitably, these led to an increase in school absences and reduced social interaction giving rise in the long-term to low self-esteem and poor sense of self-control. Recently prophylactic treatment, often administered by a parent at home, has been introduced. This has further reduced the handicap associated with haemophilia.

β-thalassaemia major is a chronic illness presenting with severe anaemia, growth retardation, hepatomegaly, splenomegaly, jaundice, skeletal abnormalities and characteristic facial appearance in later life. It requires regular blood transfusions and often regular infusion of the chelating agent desferroxamine administered overnight with a battery-powered syringe pump, 5–7 days week^{-1}. The therapeutic regime remains complex, life-long and inconvenient causing a significant burden to both children and parents. Often there is a high rate of non-compliance with treatment.

Both haemophilia and β-thalassaemia are chronic family illnesses and hence constitute a stress that

Collaborators: E. Goldman, B. Wonkie and H. Sadowski, London; T. Mandalaki, C. Kattammis, M. Piperia, S. Baharaki, D. Tsaklakidou, D. Karafoulidou, H. Berdousi and C. Samara, Athens; M. Mannucci, A. Gringeri, C. Vigano, A. Bielli and R. Berger, Milan.

Professor Israel Kolvin died following the completion of this work.

Correspondence: Dr Carmen Clemente. Child and Adolescent Psychiatry, Royal Free Hospital, Pond St, NW3 3DP, London.
Tel.: 0207 830 2931; fax: 0207 830 2810;
e-mail: clementec@noot.freeserve.co.uk

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affects not only the individual with the illness but also other members in the family who may become more vulnerable [1,2]. The possible psychosocial repercussions of chronic illness in children on the family have been researched. There is a presumption in the literature that if a family is exposed to physical adversity, there is a likelihood of increased fragility of the family's capacity for social and emotional adjustment. In a study of chronic illness [3], it was reported that children with chronic physical conditions, including haemophilia, were at increased risk of poor adjustment, compared with healthy controls.

Other factors are also likely to be important, such as the individual's coping style, family support and education, and the visibility of the illness [4]. Diseases that may be associated with limitations of physical activity, as can be the case for haemophilia, or altered physical appearance, as in β-thalassaemia are likely to have a greater negative impact on social adjustment [5]. Further, there is a belief that chronic illness itself may compromise on attachments and relationships. However, the evidence supporting such presumptions in haemophilia and β-thalassaemia is scarce, and at time contradictory. Review of research in haemophilia concluded that the psychosocial adjustment of men with haemophilia did not differ from healthy controls [6]. In a study of children who suffer from a chronic illness, haemophilia or diabetes, compared with healthy controls there was no difference between the groups expect for a small group of HIV-positive haemophiliacs who did not hold future expectations, such as getting married and having children [7].

This paper endeavours to characterize children's individual and family social adjustment when exposed to chronic physical illness as represented by haemophilia and β-thalassaemia.

According to the literature, we expected that there would be no differences in psychosocial adjustment across the three centres. We expected some differences in social adjustment between children living in families with haemophilia and those living in families with β-thalassaemia. Children affected by the illness were expected to have more social dysfunction than their siblings.

Materials and methods

The current study is part of a cross-sectional, multinational and multicentre study into the resilience of families with blood disorders. The intention was to compare the psychosocial adjustment of affected and unaffected children coming from families with haemophilia or β-thalassaemia.

Subjects

Families with haemophilia or β-thalassaemia and school-age children were recruited from the outpatients of the Royal Free and Whittington Hospitals, London, the 'Aghia Sophia' Children's Hospital, Athens and the Institute of Internal Medicine in Milan from 1994 to 1997.

The entry criteria were that one family member was affected by haemophilia or thalassaemia major, and that the family had at least one child of school age. The intention was to collect on all children. Because of limited resources, a maximum of two children of school age per family was studied. If there were more than two children in the relevant age range, then the affected child and the sibling nearest in age were selected. Seventy-five families with haemophilia and 40 with β-thalassaemia were interviewed; 33 families participated in Milan, 23 in London and 59 in Athens. Families with haemophilia were interviewed at all three centres; β-thalassaemia families were recruited only in London and Athens. Inevitably, there were more children than families and this is described next.

In all, 168 children in 115 families were studied. The families attended the centre for the purpose of research interview in London and Milan, but in Athens the families were seen on their regular time for ambulatory treatment. For geographical and staffing reasons, it was not possible to complete the whole assessment in one session. Hence, the assessment was spread over two sessions with the expectation that all patients would attend to both. For various reasons, not all patients subsequently returned to complete the assessment. Some β-thalassaemia patients travelled long distances for transfusions and whilst it was possible to interview their parents while the children were receiving ambulant medical treatment, it was not always administratively possible or psychologically sensible to undertake an assessment after the completion of their physical treatment. Further, if there was more than one child in the same family being assessed, then the assessment of the children was arranged over two sessions. If so, social adjustment was scheduled for the second – but some patients did not return for this. The net result was that social adjustment data were gathered on only 153 of the 168 children (91%). The numbers in Table 1 relate to the original full sample; however, there are minor variations in relation to social adjustment. Although the sibling groups and the group of children with β-thalassaemia contained both males and females, individuals in the haemophilia group were all males because of the sex-linked nature of the condition.

Table 1. Children's demographic data.

	Haemophilia families		Thalassaemia families	
	Children without haemophilia $n = 43$	Children with haemophilia $n = 58$	Children without thalassaemia $n = 25$	Children with thalassaemia $n = 42$
Mean age [years and months (SD)]	11.3 (3.2)	11.8 (3.6)	13.2 (2.3)	12.1 (2.10)
Gender				
Male	26 (60%)	58 (100%)	9 (36%)	20 (48%)
Female	17 (40%)	–	16 (64%)	22(53%)
Ethnicity				
White European	39 (91%)	54 (93%)	24 (96%)	40 (95%)
Asian	4 (9%)	3 (5%)	1 (4%)	2 (5%)
Others	–	1 (2%)	–	–
Schooling	$n = 42$	$n = 56$	$n = 20$	$n = 36$
None	–	5 (8.9%)	–	1 (2.8%)
Infant or junior	26 (61.9%)	31 (55.3%)	9 (45.0%)	6 (16.7%)
Secondary	15 (35.7%)	18 (32.0%)	11 (55.0%)	29 (80.6%)
Others	1 (2.4%)	2 (3.6%)	–	–

Maximum numbers in groups are listed at top of the table. Data are less for schooling.

The findings are presented for four groups of children: (i) 54 with haemophilia, (ii) 42 children from haemophilic families who were either an unaffected sibling or had parents with haemophilia, (iii) 40 children with β-thalassaemia, and (iv) their 17 unaffected siblings.

There was a less than optimum control group as 12 of the 54 (22%) children with haemophilia did not have an internal control. Almost half of these controls 20 (47%) was their siblings, the rest was children of fathers with haemophilia. In the thalassaemia group, 23 of the 40 (57%) children affected by β-thalassaemia did not have a control.

Instruments

Social adjustment Over the last two decades, there has been an increasing interest in the adjustment of patients who come into contact with medical services [8]. The assessment of social adjustment is distinct from but complements that of psychiatric psychopathology and is especially useful in the evaluation of post-treatment progress of outpatient populations. The original Social Adjustment Scale is administered by a semi-structured interview by a trained rater and has an acceptable level of reliability represented by a mean correlation of 0.83 [8–10] over all items. In a later modification, five global judgements are made on the basis of a 'community norm for social functioning' [8]. The original format was geared to adults, as some of the themes were considered inappropriate for school children (e.g. social adjustment at work or sexual relationships). However, the children were interviewed using a modified version of the Social Adjustment Scale, the questionnaire assesses social functioning in four global areas: (i) age appropriate social leisure activities, (ii) school role and relationships (rather than work role), (iii) relationship within the family and (iv) overall social adjustment.

The Social Adjustment Scale was modified for children initially by Dr Sugum-Paliwal (personal communication) and subsequently by Ms R. Miller. Piloting revealed that current adjustment was best portrayed using a 2-months-time span and past adjustment covering a 12-months-time span. In order to ensure there was adherence to the same standards of diagnostic coding across cultures, consensus checks were undertaken on data from all the three centres by a research interviewer [C-Clemente (CC)] and a senior researcher [I-Kolvin (IK)]. IK had not been involved in any of the assessments and was also blind to the patients' medical diagnostic category.

Sociodemographic data Sociodemographic data were elicited from the parents by standard semi-structured interview questionnaires previously used [11], all with satisfactory reliability. The themes included questions about the parents' accounts of their own relationship with each individual child, of their child's relationship with siblings or peers, on parental concern about their children's behaviour or emotional state, including any previous contact with

child guidance or other mental health professionals. Demographic data were incomplete for nine of the 115 families.

Socioeconomic status (SES) was determined by the Hollingshead Redlich two-factor method [12]. The main breadwinner's Hollingshead occupational codes were grouped according to three SES levels as follows: Lower Social Strata = 10–35; Middle Social Strata = 40–65 and Higher Social Strata = 70–90.

The great majority of parents (94%) were married or cohabiting. The sample included only six single-parent families (5%) – four single mothers and two single fathers – and one couple who had separated (1%). The father was the main breadwinner in 98 families (88%), and the mother in 13 families (12%). Eighteen families (17%) were coded into lower-occupational strata, 64 families (60%) middle-occupational strata, and 24 families upper-occupational strata (23%). In few families, insufficient data were available for SES coding.

The assessed children were between 6 and 18 years old with a mean age of 12 years (SD = 3.1). Children from haemophilic families were significantly younger than children from families with β-thalassaemia (t-value = 2.35, $P < 0.05$, Table 1). Children from haemophilic families were significantly younger in London than in the other two centres [F (154, 2) = 6.88, $P < 0.01$], which was true for both unaffected children [F (40, 2) = 3.47] and for the group of haemophilic boys [F (55, 2) = 3.78, $P < 0.05$]. *Post hoc* analyses showed for each comparison a significant difference between the haemophilic boys' mean age in London and in Athens (Tukey test $P < 0.05$). Thus, more children from thalassaemic families (40 of 56) than from haemophiliac families (33 of 97) attended secondary schools ($\chi^2 = 17.55$, df = 1, $P < 0.001$, OR = 4.9). While children in Milan and Athens were nearly exclusively of European or Mediterranean ethnicity, the ethnicity of the London sample included ten Asian children and one black Caribbean child.

Medical data The physical description of patients was elicited by a medical data schedule developed specifically for this study, and the medical data were collected by the patients' haematologists [2].

Severity of haemophilia was graded according to the plasma factor level in the blood: a factor level <1 U dL^{-1} indicated severe haemophilia; 1–5 U dL^{-1} moderate haemophilia, and >5 U dL^{-1} mild haemophilia. Evidence of joint damage, frequency of bleeds and school absence because of haemophilia was also recorded. For haemophilic boys, 93% ($n = 51$) had severe haemophilia and four had moderate haemophilia. Regular prophylaxis was given to 31% of the cases ($n = 17$). One-third of haemophilic boys had undergone surgery (18 of 54) at some stage of their life, and there was clinical evidence of joint damage in 39% (21 of 54). Seven haemophilic boys (13%) were HIV positive.

Severity of thalassaemia was determined according to the level of medical procedures necessary, i.e. transfusion requirements and chelation therapy, and physical complications such as bone structure abnormalities, facial appearance and growth failure. Fourteen (35%) children with β-thalassaemia had experienced at least one major complication because of their illness; 82% ($n = 33$) of thalassaemic children were undergoing various blood transfusion regimes, and all thalassaemic children (40 of 40) were treated with desferroxamine.

Clinicians were asked to rate the overall degree of their acute physical states besides the severity of the illness for all children participating in the study, on a four-point scale (no, mild, moderate or severe health problem). They rated the physical health as severely affected by their blood disorder for 82% of the thalassaemic children (33 of 40) and only 9% of haemophilic boys (5 of 54) ($\chi^2 = 47.13$, df = 1, $P < 0.001$, OR = 41.25).

Results

Psychosocial adjustment in children with haemophilia compared to children with thalassaemia

There were no significant differences between groups on social dysfunction in *leisure* settings. However, the rates appeared high in both groups with 46% ($n = 25$) in children affected by haemophilia and 40% ($n = 16$) in the group of children with β-thalassaemia. For social dysfunction at *school*, the rates were 18% ($n = 10$) in children with haemophilia and 32% ($n = 13$) in children with β-thalassaemia. Two children with haemophilia did not complete this section of the interview.

The social dysfunction within the *family* was significantly higher in the thalassaemia group, the rates in children affected by haemophilia were 28% ($n = 15$) and 47% ($n = 19$) in children with β-thalassaemia ($\chi^2 = 7.67$, df = 2, $P < 0.05$; OR = 3.9, CI = 1.6–9.3). However, for *overall* social dysfunction, the rates were similar, 43% ($n = 23$) in children affected by haemophilia and 60% ($n = 24$) for children with β-thalassaemia (Fig. 1).

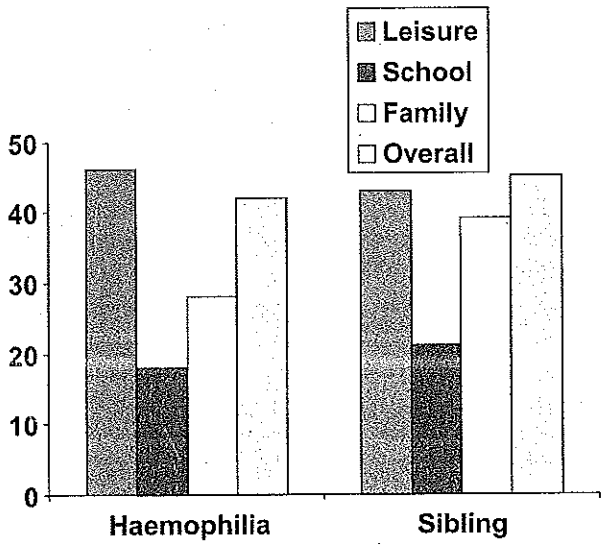

Fig. 1. Social dysfunction in children with haemophilia compared with thalassaemia.

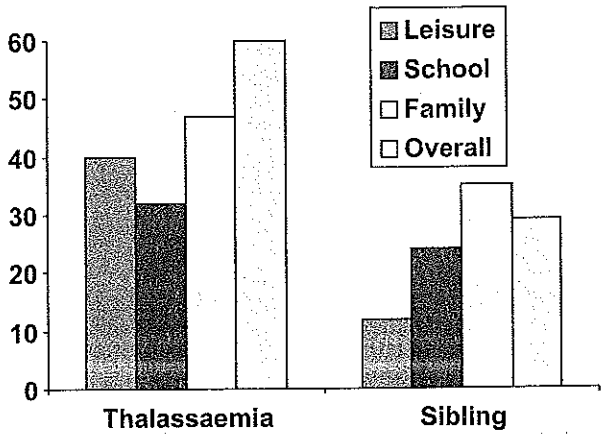

Fig. 2. Social dysfunction in children with haemophilia compared with their siblings.

Psychosocial adjustment in affected children compared with their siblings

Haemophilia. For social dysfunction in *leisure* settings, the rates for children with haemophilia were 46% ($n = 25$), which was very similar to their unaffected siblings: 43% ($n = 18$). For social dysfunction at *school*, the rates were also very similar for both groups – the boys with haemophilia had a 19% ($n = 10$) rate, and their unaffected siblings a 21% rate ($n = 9$). For social adjustment in the *family*, the boys with haemophilia had a 28% rate ($n = 15$), and their unaffected siblings, a 38% rate ($n = 16$); but this difference was not significant.

Finally, for *overall* social adjustment, the rates were 43% ($n = 23$) for boys with haemophilia and 45% ($n = 19$) for unaffected siblings (Fig. 2).

Thalassaemia Social dysfunction rates in *leisure* settings for children with β-thalassaemia were significantly higher 40% ($n = 16$) when compared with siblings without β-thalassaemia – 12% ($n = 2$, $\chi^2 = 4.7$, df = 1, $P < 0.05$; OR = 5; CI = 1–24.9).

For social dysfunction at *school*, the thalassaemic children had a 32% ($n = 13$) rate and their unaffected siblings-23% ($n = 4$). For social dysfunction in the *family*, children with β-thalassaemia had a 47% ($n = 19$) rate, unaffected siblings – 35% ($n = 6$); the differences were not significant. Finally, the *overall* social dysfunction rates were 60% ($n = 24$) for thalassaemic children and 29% ($n = 5$) for unaffected siblings living in families with thalassaemia ($\chi^2 = 4.1$, df = 1, $P < 0.05$; OR = 3.6; CI = 1.1–12.2).

In order to exclude cross-cultural factors, the thalassaemic data from Athens were reanalysed on its own. Sixty-six per cent (23 of 35) of those children with β-thalassaemia had significantly higher rates of *overall* social dysfunction than their unaffected siblings, where the rate was 14% (two of 14) ($\chi^2 = 7.2$, df = 1, $P < 0.01$; OR = 5.5; CI = 1.4–22.1). The above findings provide support for the notion that children with β-thalassaemia are at risk of social dysfunction.

In summary, there were no differences between the children affected by haemophilia and their siblings; however, for thalassaemia, the picture was rather different: children affected by β-thalassaemia had significantly more social dysfunction in *leisure* situations and their *overall* level of social behaviour than their unaffected siblings (Fig. 3).

Psychosocial adjustment according to age and gender

Secondary-school-age children with either haemophilia or β-thalassaemia showed significantly more social dysfunction than younger children, particularly in the area of *school* functioning (older than 11 years, 32% ($n = 32$) vs. younger than 11 years, 4% ($n = 4$), ($\chi^2 = 10.0$; df = 2, $P < 0.02$; OR = 5.2, CI = 1.7–15.8). There were no significant differences in the *overall* social adjustment by *gender* with 41% ($n = 43$) of boys and 57% ($n = 28$) of girls showing at least some social dysfunction. In summary, older children showed more social dysfunction at *school*. There were no differences by gender.

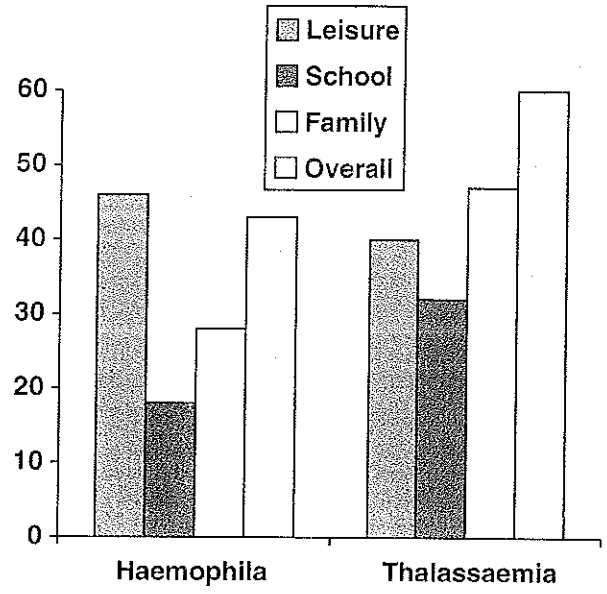

Fig. 3. Social dysfunction in children with thalassaemia compared with their siblings.

Discussion

The hypothesis that children with β-thalassaemia would show more social dysfunction when compared with haemophiliac boys was confirmed by the significantly higher level of social dysfunction within the *family in* the thalassaemia group. In a retrospective study of 28 patients with thalassaemia intermedia, 28% of the patients' social activities were reduced to a severe degree [15]. This is similar to our sample where 22% (nine of 40) of children affected by β-thalassaemia showed moderate to marked overall social dysfunction (Fig. 3).

Further, in the current study almost half 48% (19 of 40) of thalassaemic children experienced some dysfunction when compared with just over a quarter, 28% (15 of 54) of children affected by haemophilia. This figure is again similar to that reported in a retrospective study of 28 patients with thalassaemia intermedia: 51% of parents of these children described them as suffering from social isolation [14]. However, the hypothesis that children affected either by haemophilia or β-thalassaemia would show higher levels of social dysfunction than their healthy siblings was confirmed only for thalassaemic families. On the contrary, there were no differences between children with haemophilia and their siblings. It needs to be emphasized that there was no external control group in the present study. Thus, conclusions can only be drawn about affected and unaffected siblings. However, in all three centres, the rates of social dysfunction in the affected children appeared much higher than the rate expected in normal population.

Rates of social dysfunction averaged about 45% in the current study – some three times the expected rate for the normal population [13]. Much of the identified social dysfunction was at the lower levels of severity and therefore not likely to be greatly handicapping to the child; severe social dysfunction affected only a small percentage of patients in this study with the exception of the *overall* social adjustment of children with β-thalassaemia where the rate was one in four. On average, it seems wiser to continue to accept the view [15] that the siblings of chronically ill children are at risks for social dysfunction especially when there is physical deterioration in the index child.

On theoretical grounds, it is to be expected that healthy siblings of children affected by a blood disorder would be at risk of psychological dysfunction for a number of reasons. First, their parents may be devoting what to the unaffected siblings appears to be an excess of time, care and affection to the child affected by a blood disorder. Some anecdotal case studies and clinical research supports the notion that siblings may resent what they perceive as this special attention and preferential treatment being given to a chronically ill child. While family dynamics may prevent the emergence of any overt hostility or jealousy, repressed hostility may reveal itself in a number of ways including deterioration of schoolwork [16]. However, if the illness has a genetic basis, the presence of healthy siblings may have a positive effect, in that it can reduce parental feelings of guilt and inadequacy [17]. While on balance, the available evidence suggests that siblings of patients with chronic illness are at risk of psychological disturbance and interpersonal problems, more recent epidemiological research does not consistently support these conclusions.

Siblings of the chronically ill enjoy generally good mental health and social well being [13]. They described only the modest tendency to emotional problems and speculated that some of these identified problems might be a consequence of an expectation bias by the assessing clinicians. In contrast, they reported that children with the chronic illness tended to have a twofold increase in psychiatric disorders compared with their peers, but only a small excess of social adjustment problems. If the chronic illness was associated with limitations of physical function, the affected children then had a threefold increase in

psychiatric disorders, particularly a significant excess of social adjustment problems.

The presence of higher rates of medical problems in the thalassaemic group could explain the high levels of social dysfunction in *leisure*. These medical problems were significantly higher than in the haemophilia group, e.g. 81% of thalassaemic needed regular blood transfusions.

A possible additional important contributory factor to more severe social dysfunction in the thalassaemic group was that 95% of them were receiving subcutaneous desferrioxamine infusions for up to 10 h for five to seven nights per week. In contrast, the treatment received by the haemophilia group was less burdensome with only 31% of cases receiving regular prophylaxis and that being much less burdensome. Thus these medical treatment differences could well explain some of the between group differences for social dysfunction in *leisure*. This hypothesis is reminiscent of the views emerging from the Ontario Health Study findings [13] where children with chronic illness associated with limitations of physical function, had a threefold increase in social adjustment problems.

In *summary*, children living in a family with a blood disorder showed an excess of social dysfunction higher than expected. Most of this dysfunction was mild rather than moderate to marked. Children from families with haemophilia whether affected or not had double the expected rate of social dysfunction as reported in a general population study irrespective of whether they were unaffected siblings or had the condition [13]. Rates for affected and unaffected children did not differ from each other. Children with β-thalassaemia showed three times the population expected rate of social dysfunction. This could have multiple determinants; including the disorder itself; the consequences of more intrusive and complex forms of medical treatment procedures; a psychological reaction to the bodily changes; and inadequate educational experiences.

There are some important limitations. First, some of the defined cohorts had small sample sizes and so caution is necessary in interpreting this data. Secondly, this study is one of group comparisons of affected children with their unaffected siblings. While this method provides some answers to crucial clinical questions, there are questions remaining about how the rates in the various subsamples compare with what might be found in a general population study. Unfortunately, there were insufficient resources to address this theme but clearly, it merits further investigation.

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