ORIGINAL ARTICLE

Coping and health service utilisation in a UK study of paediatric sickle cell pain

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Aims: To assess sickle cell pain and coping in children and to examine the relation between these factors and the utilisation of health services.

Methods: Cross sectional study involving 67 children with sickle cell disease attending three London hospitals. Interviews and questionnaires involved measures of pain, health service utilisation, and coping responses (measured with the Coping Strategies Questionnaire (CSQ), revised for children with sickle cell disease). Medical data on complications, haemoglobin (Hb) levels, and foetal haemoglobin (HbF) percentage were also collected.

Results: Pain accounted for about 24% of hospital service use, independent of age, sex, number of with sickle cell disease complications, and Hb levels. However, 42% of patients had not utilised hospital services in the past 12 months. Three higher order factors emerged from analysis of the CSQ (active coping, affective coping, passive adherence coping). Pain severity was predicted by passive adherence coping, while utilisation of hospital services was predicted by active coping.

Conclusions: Sickle cell disease in children involves severe recurrent pain leading to hospitalisation in some cases. Psychological coping patterns are relevant to both pain experience, and the use of acute hospital services. It is likely that children would benefit from community based interventions that incorporate both medical and psychological assessments.

About 170 babies are born annually in the UK with sickle cell disease (SCD),[1] a group of genetic conditions resulting from abnormal haemoglobin synthesis. SCD includes sickle cell anaemia (HbSS), sickle haemoglobin C disease (HbSC), and sickle β thalassaemia disease (HbSβThal). In the UK, SCD mainly affects people of African and Caribbean origin, but is also found in people originating from the Mediterranean region, southern Europe, the Middle East, and Asia. Recurrent pain is the predominant symptom associated with SCD, resulting from the occlusion of blood vessels by "sickle shaped" red blood cells, usually referred to as "sickle cell crisis". Other acute and chronic complications can occur, including splenic sequestration, sickle acute chest syndrome, organ damage,[2] and an increased risk of stroke in children.[3] Life expectancy has dramatically improved through better understanding and advances in medical treatment.[4] Despite substantial progress, young people with SCD still suffer many health related problems and social disadvantages.[5 6] Pain and other complications associated with SCD may begin in infancy and persist throughout life, affecting an individual's quality of life. It is therefore important to identify coping strategies used in response to pain during childhood as these may remain through adulthood. It is also imperative that both medical and complementary management (including education, support, counselling, and specific psychosocial interventions) of SCD is aimed at improving the quality of life of patients.

Previous studies in the USA have found that psychologically passive coping responses and negative thought patterns were associated with pain experience and health care utilisation in children and adolescents with SCD.[7 8] These studies also suggest that pain, adaptation, and health service utilisation are related to psychological coping responses independently of SCD severity. Work in children using similar measures has not been carried out in the UK, where the different structure of health services may lead to different findings. We therefore aimed to examine the relations between pain, coping, and health service utilisation in a UK sample, and determine whether these bear any similarities to previous findings in the USA. We hypothesised that pain experience and utilisation of health services in sickle cell children are influenced by their coping strategies, independently of clinical condition.

METHODS

Patients

Sixty seven children aged 7–15 years with SCD, attending St George's, King's College, and Central Middlesex Hospitals in London took part in the study. These children were all English speaking, and community based at the time of interview. The aim was to recruit 65 children for the planned factor analyses. A total of 84 parents of sickle cell children were contacted over a time period of six months for each hospital; nine of them refused consent for their child to participate. Various reasons were given for refusal, including lack of interest in research, no medical benefit, and no time. None of the children were excluded for inability to understand English. Time constraints did not allow the remaining eight children to be interviewed. The age range of the children was consistent with previous research using similar measures.

Measures

Pain status and health service utilisation

Pain and health service utilisation were assessed using a pain interview that records parents' reports of the number and duration of pain episodes within a 12 month period preceding the interview, and children's ratings of pain severity. This interview is based on the standard pain interview developed for adult sickle cell patients.[9] Parents reported on their child's

Abbreviations: CSQ, Coping Strategies Questionnaire; Hb, haemoglobin; HbF, fetal haemoglobin; HbSαThal, sickle α thalassaemia disease; HbSβThal, sickle β thalassaemia disease; HbSC, sickle haemoglobin C disease; HbSS, sickle cell anaemia; SCD, sickle cell disease

Table 1 Demographic and clinical characteristics of sickle cell children

Variable	St George's Hospital (n=22)	King's College Hospital (n=23)	Central Middlesex Hospital (n=22)	Total (n=67)
Age*	10.9 (2.2)	11.6 (2.3)	11.5 (2.0)	11.3 (2.2)
Hb level (g/l)*	84 (10)	83 (15)	94 (26)	87 (18)
HbF (%)*	4.6 (4.0)	3.6 (2.3)	6.6 (3.3)	4.8 (3.4)
Complications*	0.5 (0.7)	0.5 (0.8)	0.6 (0.9)	0.5 (0.8)
Gender	8 (37%)	11 (48%)	11 (50%)	30 (45%)
Female	14 (63%)	12 (52%)	11 (50%)	37 (55%)
Male				
Phenotype	15 (68%)	14 (61%)	16 (73%)	45 (67%)
HbSS	5 (23%)	7 (31%)	4 (18%)	16 (24%)
HbSC	2 (9%)	1 (4%)	2 (9%)	5 (7%)
HbSβThal	0 (0%)	1 (4%)	0 (0%)	1 (2%)
HbSαThal				
Ethnic origin	13 (59%)	19 (83%)	9 (41%)	41 (61%)
African	7 (32%)	4 (17%)	12 (55%)	23 (34%)
Caribbean	2 (9%)	0 (0%)	1 (4%)	3 (5%)
Mediterranean				

*Results expressed as mean (SD).

health service utilisation within a 12 month period preceding the interview. Health service utilisation was measured only in terms of medical care. Visits to accident & emergency departments, the number and duration of hospital admissions, and consultations with their general practitioner for pain were ascertained. Types of pain analgesia taken at home and treatment in hospital were also recorded. Questions included: "How many sickle cell pain episodes has your child had in the past year?" and "How many times has your child been hospitalised because of sickle cell pain in the past year?".

Psychological coping

The Coping Strategies Questionnaire revised for SCD children (CSQ-SCD)[7] was used to assess behavioural responses and cognitive strategies used by patients to manage and control sickle cell pain. This self-complete questionnaire assesses several general ways of coping with pain (including diverting attention, reinterpreting pain sensations, calming self statements, ignoring pain sensations, praying and hoping, catastrophising, and increasing activity level), together with six scales developed as specifically relevant to SCD (fear self statements, anger self statements, isolation, taking fluids, resting, and heat/cold/massage). Children rated their use of each response from 0 = never to 6 = always; scores were averaged to produce a mean for each scale.

Medical records

Medical records of the children were examined for markers of disease severity. These were complications associated with sickle cell crises, steady state haemoglobin level (Hb), and most recent recording of fetal haemoglobin (HbF) percentage.

Procedures

Lists of sickle cell children registered at three hospitals (St George's, King's College, and Central Middlesex Hospitals) were obtained from the haematology departments. Children were recruited opportunistically through outpatient clinics in all three hospitals. Those who were not seen at the outpatient clinics were pursued in the community by means of a letter written to their parents. Interviews were arranged at a suitable time and location (usually in the child's home) and were conducted in the presence and with the consent of a parent, usually the mother. The pain interview was administered to parents, but children rated pain severity and completed the CSQ-SCD.

Statistical methods

Correlations and *t* tests were performed initially to determine variations in demographic and disease severity factors of age, gender, phenotype, ethnic origin, Hb level, HbF percentage, number of complications on measures of pain, health service utilisation, and coping. Furthermore, the associations between pain, health service utilisation, and coping were analysed using hierarchical stepwise regression. In each analysis, demographic and clinical variables (age, sex, phenotype, number of complications, Hb) were entered on the first step. On second and subsequent steps, the independent variables were entered with the forward method according to their strength, with $p < 0.05$ being the criterion for entering. We report the additional variance accounted for, together with the F change, for each step.

RESULTS

Demographic and clinical characteristics

Table 1 shows the demographic and clinical data of 67 children who participated in this study. Four phenotypes were identified, comprising 45 children with HbSS, 16 with HbSC, five with HbSβThal, and only one with HbSα thalassaemia disease (HbSαThal). There were no significant differences between the three main phenotypes (HbSS, HbSC, HbSβThal), gender, age, and ethnic origin on measures of disease severity ($p > 0.05$).

Pain status

Descriptive data of the pain measures were compared with data reported in a previous US study[7] (table 2) involving children and adolescents of similar age (7–17 years). The number of pain episodes over the past year reported for sickle cell children in this study was significantly lower than the numbers recorded in the earlier study ($t = 3.44$, $p < 0.001$). However, pain duration and severity measures did not show any significant differences between the two studies. Pain severity ratings among children of three phenotype groups in this study (HbSS, HbSC, HbSβThal) differed significantly. Children with HbSS reported more intense pain than the others ($F = 4.06$, $p = 0.022$).

Health service utilisation

Table 2 shows the mean levels of health service utilisation. No significant differences were found in the number of emergency visits and hospital admissions between sickle cell children in this study and those reported in a US study.[7] A small proportion of sickle cell children made the majority of contacts with health services in the previous year. Thus, 61

Table 2 Pain and health service utilisation, in comparison with a study in the USA

	Present study (n=67)	Gil *et al* (1991)* (n=72)
Number of pain episodes (12 months)	3.8 (5.3)	8.0 (8.6)
Duration of pain episodes (hours)	98.0 (114.6)	83.5 (88.8)
Pain severity rating	7.3 (2.3)	6.6 (3.1)
Emergency visits (12 months)	1.4 (1.8)	1.4 (1.8)
Hospital admissions (12 months)	1.4 (1.7)	1.0 (1.7)
Duration of admissions (days)	4.7 (6.8)	
General practitioner visits (12 months)	1.4 (2.2)	

Results expressed as mean (SD).
*Gil *et al*.[7] Nine month data scaled up for 12 month comparisons.

Table 3 Average scores for coping scales, and higher order factor analysis

Psychological coping strategy	Mean (SD)	Factor 1	Factor 2	Factor 3
Active coping				
Ignoring pain sensations	2.21 (1.44)	0.84		
Calming self statements	3.04 (1.40)	0.81		
Increasing activity	2.63 (1.24)	0.80		
Diverting attention	2.61 (1.38)	0.80		
Reinterpreting pain sensations	1.58 (1.35)	0.74		
Praying and hoping	3.49 (1.32)	0.51		
Affective coping				
Catastrophising	2.77 (1.37)		0.87	
Anger self statements	2.50 (0.92)		0.87	
Fear self statements	2.75 (1.26)		0.79	
Isolation	2.23 (1.39)		0.52	
Passive adherence coping				
Taking fluids	4.34 (1.33)			0.70
Resting	4.29 (1.09)			0.70
Heat/cold/massage	2.97 (1.07)			0.57

(66%) accident & emergency visits were made by 11 (16%) patients, while 58 (63%) hospitalisations occurred in 14 (21%) of patients (not the same). Similarly, 63 (64%) general practitioner visits were carried out by 11 (16%) patients. Interestingly, 42% of children had no hospital admissions and 60% had no consultations with their general practitioner in the 12 months prior to the interview. Hospital service indices were not associated with visits to the general practitioner. No associations were found between demographic or clinical variables and health service utilisation.

Intercorrelations between the number of emergency visits and hospital admissions was significant ($r = 0.76$, $p < 0.001$), and a "hospital service utilisation index" was constructed for sickle cell children by adding together these two measures. A hierarchical regression analysis on the hospital service utilisation index was performed. Clinical and demographic factors (phenotype, age, sex, number of complications, Hb) were entered first, followed by the pain measures in a forward stepwise fashion. The clinical and demographic variables accounted for only 3.6% of the variance in the hospital service utilisation index ($F = 0.413$, $p = 0.838$). Pain frequency and severity were independent predictors, and together accounted for an additional 23.8% of the variance ($\beta = 0.241$ and 0.433 respectively, F change = 10.9 and 5.9, $p < 0.02$). A second regression on general practitioner visits was carried out, but none of the pain measures entered the model. These findings indicate that the use of acute hospital but not general practitioner services in sickle cell children can be predicted by pain experience after demographic and clinical factors have been taken into account.

Psychological coping

The CSQ-SCD for children was subjected to higher order factor analyses to determine the extent to which the various coping responses were related to each other. Three factors had eigen values >1.5 and together accounted for about 63% of the variance. The varimax loadings on the three factors described show high internal consistency (table 3). Factor 1 was defined "active coping": sickle cell children scoring high on this factor seem to cope with pain actively using cognitive and behavioural strategies (Cronbach $\alpha = 0.93$). Factor 2 was defined "affective coping": sickle cell children scoring high on this factor employed negative thinking and emotional responses, including catastrophising, self statements of fear, and anger. The Cronbach α for this factor was 0.87. Factor 3 was defined "passive adherence coping": sickle cell children scoring high on this factor appeared to utilise pain coping methods recommended by doctors, such as increasing fluid intake and resting, rather than active psychological techniques. The Cronbach α for this scale was 0.76. These three factors were utilised in the subsequent analyses. Clinical and demographic variables were not associated with coping.

Coping and pain

Separate hierarchical regression analyses were used to determine the associations between the three coping factors and pain measures. In each case demographic and clinical variables were entered in step 1, followed by the three coping factors in a forward stepwise method. None of the coping factors contributed to the regressions on the number or duration of painful episodes. However, in the analysis of pain severity, the passive adherence coping factor entered the model after demographic and clinical variables ($\beta = 1.112$, F change = 10.15, $p = 0.002$). Pain severity is predicted by passive adherence coping, which accounts for an additional 15.5% of variance. Children who use more passive adherence coping methods experience more intense pain.

Coping and utilisation of health services

Regression analysis of the hospital service utilisation index entered demographic and clinical data in the first step, followed by the three coping factors. Active coping accounted for an additional 16.4% of the variance, indicating that children who respond to sickle cell pain with active coping responses, such as increased activity, are more often found in hospital than other children ($\beta = 0.337$, F change = 3.13, $p = 0.035$). This is not a result of greater pain as active coping was not significantly associated with any of the pain measures. In addition, when pain frequency and severity were included in the model, the active coping factor accounted for 9.4% additional variance ($\beta = 0.318$, F change = 6.84, $p = 0.012$). The other two coping factors did not make a significant contribution to the variance in the hospital service utilisation index. There were no associations between coping and general practitioner visits.

DISCUSSION

This study forms part of a series of studies with sickle cell children and adults attending hospitals in London. Many children were found to be prone to vaso-occlusive pain episodes and associated complications. The analyses indicate that sickle cell pain on its own did not account for the number of hospital admissions or the length of stay in hospital. A number of children were admitted to hospital because of complications. One common reason for the onset of a painful crisis, as well as hospital admissions and extended hospitalisations in children, is infection. Children at risk for a stroke are also often treated with a transfusion regime in hospital, although these transfusions are usually on a day care basis. Other measures of disease severity, namely Hb level and HbF percentage, are of particular importance in sickle cell children. The HbF percentage usually decreases with age, and it has been shown that children with a high HbF percentage tend to experience less frequent pain.[10]

In this population, children with HbSS experienced more severe pain compared to those with HbSC or HbSβThal. This is consistent with the notion that sickle cell anaemia is the more severe phenotype of the condition.[11] Gender and age (unlike previous research[7]) differences were not found to influence pain status. By comparison, children in this study had significantly less frequent pain than those in a previous US study.[7] This may be attributed to our community focused approach, whereby health professionals (including specialist nurses and psychologists) actively follow up children who make little or no contact with hospital services at home. No significant differences were found in pain duration and severity reports of the two studies, indicating that these aspects of sickle cell pain experience are similar in the UK and USA.

The health service utilisation data indicate that many children with SCD have little contact with health services in an average year; 42–45% had no accident & emergency visits or hospital admissions, and 60% had no contact with their general practitioner in a year. Other problems apart from pain may lead to the utilisation of health services. The effects of symptoms in sickle cell children and the quick response to the treatment of complications are of primary concern to haematologists. Thus, utilisation of health services is not likely to be a cause of anxiety among doctors. However, clinical factors do not entirely account for all the reasons why children utilise health services. Pain frequency and severity in this study predicts a significant proportion (24%) of the variance in hospital utilisation beyond that accounted for by demographic and clinical variables. Consistent with previous reports,[7 8] children who had more frequent and intense pain also made more contacts with hospital services.

The use of hospital services was not associated with general practitioner consultations. This finding is interesting, and it seems that sickle cell children use primary and secondary health services differently. Some patients go to their general practitioners when they are in pain, while others avoid primary care to use hospital services. The factors that determine these preferences have to be carefully examined, given that continuity of care within the community is crucial for sickle cell children. Nonetheless, it is possible that patient satisfaction with the skills and knowledge of general practitioners is relevant.

Our underlying hypothesis was supported. In addition to pain, coping responses were found to predict both pain experience and the utilisation of health services. Passive adherence coping was positively associated with pain severity. These responses, such as resting and taking fluids, may augment pain, and although they are physiologically important, and are advised by physicians, may not be sufficient in controlling pain. Possibly, these psychologically passive strategies also make children feel helpless and out of control, and thus have a negative effect on pain experience. On the other hand, some children may have so much pain that they do not feel able to do anything other than employ these passive behaviours. By contrast, active coping responses were associated with hospital utilisation. Children using active coping responses also utilise acute hospital services more, but not because they have worse pain. Parents are usually advised to take their children to the hospital when they are unsure how to manage the symptoms, or when there is any suspicion of complications. Hence, the decision to utilise health services is not made by the child and may not necessarily be a result of pain. Also, it might be the case that the use of hospital services could be seen as another aspect of active coping. If someone takes a problem solving approach to their illness, this might include active use of health services along with other painful behaviour. Though active coping responses may not influence pain, once there is an episode, children may use these methods to cope better.

Our findings relating coping with health service utilisation are in contrast with previous work,[7 8] in which children with SCD who used more active coping responses utilised hospital services less. One possible explanation could be our focus on a community based sample whose coping methods are perhaps different from those of patients studied in hospital. It may also be the case that parents determine health service utilisation irrespective of a child's coping responses. These suggestions could be explored further with qualitative analyses of the reasons patients and their parents give for seeking medical support. Contrary to earlier studies, demographic and clinical factors such as age and phenotype cannot predict health service utilisation and coping.

There were some limitations to the study. The cross sectional design made it impossible to tease out causality, which was influenced by the lack of longitudinal data. The opportunistic selection of children for the study may have been biased; however, a community sample was obtained representing three areas of London. Measures of pain status and health service utilisation were retrospective, as is true in much pain research. Much of the information was supplied by parents instead of children, in order to increase the chance of obtaining accurate reports of health service utilisation. It is possible that children's own accounts would have been different. In addition, there was no opportunity for measuring parental coping responses, but these may be important, and could have influenced those of their child. Nonetheless, the study contributes to our understanding of sickle cell children's coping and health service utilisation in the UK.

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