PSYCHOSOCIAL BURDEN OF SICKLE CELL DISEASE ON CAREGIVERS IN A NIGERIAN SETTING

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In Nigeria, with a high prevalence of sickle cell disease (SCD), families bear most of the burden of care for patients with this chronically disabling illness, because there are no national social welfare provisions. To assess the severity of indices of psychosocial burden among relatives of 24 SCD patients in crisis, in comparison with relatives of 57 SCD patients in stable condition. Cross-sectional study, using a burden instrument and Goldberg's General Health Questionnaire. Objective burden indices were significantly higher for SCD in crisis. The financial burden of SCD in crisis was significantly higher than the burden of disruption of family routines. 57.9% of all caregivers experienced little or no difficulty coping with SCD. Relatives with higher educational and occupational attainments (compared with those with lower attainments) experienced significantly lesser financial burden, disruption of family routines, and psychological distress. Compared with a similar study of cancer patients, relatives of patients in SCD crisis perceived similar financial, family routine burdens and psychological distress scores. Compared with US and UK reports, our caregivers were predominantly married and reflected the national range of socioeconomic groups. Global rating of burden was significantly predicted by disruption of family routines and higher age of caregivers. The psychosocial burden of SCD can be significantly reduced by controlling the frequency and duration of crises, as well as providing adequate information and socioeconomic support to families. Hematology staff should be sensitive to the psychosocial dimensions of SCD. (*J Natl Med Assoc.* 2002;94:1058–1070.)

Key words: sickle cell disease ◆ psychosocial ◆ burden ◆ family ◆ Nigeria

INTRODUCTION

Sickle cell disease (SCD) is a group of blood disorders, including sickle cell anemia (HbSS), sickle cell hemoglobin C disease (HbSC) and sickle cell B Thalassaemia (SB Thal), which have in common a tendency for red blood cells (RBCs) to sickle (or distort into a crescent shape) under certain conditions. The deformed RBCs create blockages in small blood vessels, leading to oxygen deprivation and tissue damage. The medical symptoms associated with SCD appear to stem from the disruption of normal circulation caused by sickled RBCs. A

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sickle cell crisis is typically a recurrent episodic phenomenon, lasting several days, characterized by severe pains, fever and debilitation. Pain crises are one of the common causes of distress among patients with SCD,[1] and account for 90% of SCD-related hospital admissions.[2]

SCD afflicts up to 100 million people worldwide, predominantly black people (in Africa, Europe and the Americas), Arabs, and those of Asian ancestry.[3] In Nigeria, the incidence of HbSS is about 2% and that of HbSC is approximately 0.7%.[4] The life expectancy of patients with SCD is quite variable. Some die at an early age while others have a virtually unrecognized condition and are able to live active lives; only a few live until advanced age. In the US, it is estimated that 50% of SCD subjects survive beyond the fifth decade,[1] while in Nigeria, an increasing number of patients are now surviving beyond the age of 30 years. Progress in the scientific knowledge of SCD and community awareness programs have made it possible for patients to have access to treatment techniques that have helped many of them to live longer. With this achievement, attention is now focusing on the psychosocial dimensions of the illness, in order to achieve a better quality of life for SCD subjects and their families.

From the decade of the 1980s, there has been an upsurge of research focusing on the psychosocial aspects of SCD. Midence and Elander,[5] in reviewing these works, have noted that the majority of the studies have been based on US samples, with an increasing number from Britain; and that Nigerian researchers have led in the field outside of the Western world. Earlier Nigerian reports were anecdotal,[6] highlighting the possible psychological impact of SCD. In the 1990s, Nigerian researchers used standard questionnaires to screen for mental disorders among SCD subjects,[7,8] results of which indicated that about a quarter of clinic attendees have clinically significant psychological symptoms. The attendees commonly worried about the limitations that the illness placed on social life, depressive feelings, abnormal body shape, suicidal ideation during crises, and the burden of illness on the family.[9]

As evidence of the wider social impact of the disease, social scientists working in Nigeria investigated whether SCD sufferers were traditionally labeled as "Ogbanje" (among Igbos) or "Abiku" (among Yorubas). The Ogbanje/Abiku phenomenon is a reincarnation theory among the Igbos and Yorubas of southern Nigeria, concerning persons who are believed to be rapidly cycling into brief periods of earthly existence.[10] In this theory, if a family loses a number of preadolescent children, it is believed that the death is caused by a single soul who was predestined to have a succession of short lives. Researchers found that the use of the term Ogbanje/Abiku may be inclusive of, but not limited to SCD.[11]

In Nigeria, the tendency for SCD subjects to become dependent on analgesics has been highlighted in a case report,[12] which is in line with our ordinary clinical experience.

Although these Nigerian workers commented on the possible psychosocial burden of SCD, the only report that specifically focused on caregiver burden was limited to articulating a questionnaire for assessing burden on the family of SCD sufferers.[13] In a computer search of the scientific literature, we could not find any other reports from Africa in which the psychosocial burden of SCD on caregivers was systematically assessed.

Hill[14] and Midence and Elander,[15] in reviewing the literature on caregiver burden from the US and UK, have shown the salutary effect of psychosocial support and social welfare programs on the family's adjustment to the problem and management of SCD. A common finding was that SCD subjects were acutely aware that their illness was a burden on their families.[16] Because of the chronicity and severity of SCD, caregivers must guide patients in the demanding daily management of this condition.[17] When pain episodes occur, caregivers assist in managing pain by offering physical and emotional comfort, providing analgesia, or coaching the patients in pain-coping techniques

(e.g., distraction, relaxation). For more severe pain or symptoms that may signal other physical complications, caregivers must seek medical attention for the patients.

In Nigeria and most sub- Saharan African countries, the increasingly fragile extended family system has to bear the major burden of chronic medical illnesses, because there are no national social welfare provisions.[18] However, the issue of the impact of chronic illness on social networks in sub-Saharan Africa is neglected. Hence, there is a need for a systematic assessment of caregiver burden, with a view to highlighting the types of support that require priority attention for policymakers, lay self-help groups and philanthropic bodies. In this regard, some of the basic research questions that could provide useful information are: (1) To what extent have the family's finances, as well as economic and routine home activities, been affected by caring for the patient? (2) How has caring for the patient affected the emotional atmosphere in the family and relationship with neighbors in the community? (3) How much psychological distress - as measured by the number of non-psychotic psychiatric symptoms - is associated with the caregiving role? (4) How would caregivers rate the overall (global) difficulty of caring for the patient? What are the factors associated with this rating? And how does this rating compare with the ratings of caregivers of patients with other chronic debilitating physical illnesses in the same locality? (5) Is the perception of burden related to whether the patient is in crises (i.e., is burden state-dependent)?

In the literature pertaining to burden, these questions have been pursued, either by a qualitative method in which the detailed experiences of a few caregivers are described,[14] or by a statistical analysis of quantitative data.[13,17] The method chosen for the current study was quantitative analysis of data obtained by interviewing caregivers.

The general aim of the study was to assess the severity of various indices of psychosocial burden experienced by family caregivers of SCD patients attending the hematology outpatient clinic of a Nigerian general hospital, and the factors associated with burden. The specific objectives were: to highlight (1) the extent to which caregivers have lost financial benefits or have been prevented from earning income because of their caregiving roles; (2) how much the expenditure incurred due to patient's illness has affected the family's finances; (3) the extent to which caring for the patient has caused disagreements or quarrels among family members; (4) whether the family feels socially stigmatized because of having an SCD member; (5) the types of non-psychotic psychiatric symptoms experienced by caregivers, using a simple psychiatric screening instrument; (6) the overall (global) rating of difficulty of coping with the patient's condition; (7) the differences between the responses of relatives of SCD patients who presented as emergencies (i.e., in crises) and those of relatives of patients in stable condition (i.e., in a steady state, having been without crises for at least six months) who attended for routine care; (8) the factors associated with a global rating of difficulty of coping with a patient.

We were interested in comparing the two groups of SCD caregivers because we wanted to see whether the effect of currently caring for a relative in crisis would differentially affect caregiver perception of burden after controlling for possible differences in disease severity. The results are compared with the findings of a recent similar study of cancer patients in the same hospital[19] and discussed in the light of similar studies from developed countries.

METHOD

Burden Questionnaire

Our research used a cross-sectional study and our operational definition of "burden" followed that of Platt,[20] namely, the presence of problems, difficulties, or adverse events which affect the lives of patients' significant others (e.g., members of the household and/or the family).[19] In particular, burden was taken to

refer to that element of hardship explicitly attributed to the patient.[21]

To determine caregiver burden for cancer patients, our center uses a successfully field-tested, Nigerian culture-relevant instrument, which could be easily adapted for studying other chronic medical illness groups by inserting the appropriate disease name.[19] The instrument was articulated by us for psychosocial studies of caregiver burden in different medical conditions in our setting, and therefore, did not require elaborate standardization for study of SCD caregivers in our locality. Based on our experience of psychosocial problems of SCD in the country,[9] the instrument was accordingly adapted for the present study of SCD caregivers, after a validity exercise.

The contents of the instrument were guided by the domains of burden highlighted by Platt[20] and Hoening and Hamilton.[22] Therefore, items of the instrument were constructed to elicit responses regarding the severity of "objective" and "subjective" burden.[22] Under objective burden, we recorded a broad range of adverse effects on the family, such as financial loss; impairment of work efficiency; disruption of routine family activities (e.g., household work and attention to other family members); disruption of family interactions (e.g., emotional climate at home, leisure, relationship with outsiders); and physical health. Under subjective burden, we recorded what the relatives themselves felt about the illness, and the general feeling of difficulty they experienced in caring for the patient (details of the questionnaire are available from the authors upon request).

Two typical items to elicit responses on key domains of objective burden are as follows:

A. Has the patient lost any sort of revenue or financial benefits (e.g., loss of job, loss of working hours for business or going to farm or going to sell at the market, etc.)?
   -No such losses / Minor loss of revenue or benefits / Moderate loss of income / Major loss of income, which has greatly affected the family
B. Has taking care of the patient caused neglect of the attention that should be given to other members of the family (e.g., spouse leaving children at home in order to stay in hospital with the patient or take patient to other places of care, etc.)?
   -Not at all / occasionally / Frequently / All the time

The items pertaining to objective burden, therefore, involved four response options with increasing severity as above (each item, score of 0–3) This section consisted of 12 items, with a possible maximum score of 36.

Two typical items to elicit responses on subjective burden are as follows:

A. Does the family feel embarrassed that so many people have to know that the patient has this sickness?
   -Not at all / Occasionally / Regularly
B. Global rating of burden was assessed by the following question: On the whole, how much difficulty do you have coping with this patient's condition?
   -None at all / Mild difficulty / Moderate difficulty / Severe difficulty

This section of the instrument consisted of eight items, with a maximum score of 24. In assigning scores, the ideas of "frequency" and "severity" were used interchangeably, depending on the measure.

The first part of the instrument contained items about socio-demographic characteristics of the patient and respondent. Disease severity was assessed on a longitudinal dimension by collecting information on frequency of hospital attendance and hospital admissions, frequency of severe pain, and how frequently the patient was taken to Pentecostal churches for "healing," and to traditional (native) healers for treatment.[13,17] In Nigerian culture, an indication of how serious relatives consider an illness

to be is reflected by whether the patient is taken to Pentecostal churches and traditional healers for "healing."[13]

The last part of the instrument consisted of the 12-item version of Goldberg's[23] General Health Questionnaire (GHQ-12). The GHQ-12 is a screening instrument used to assess probable psychiatric cases. It is particularly useful for physically ill subjects, because it does not contain somatic symptoms, which may be caused by the physical illness. The GHQ has been used in our locality.[24,25]

The reliability of the burden instrument when used for relatives of cancer patients has already been reported.[19] In the field trial stage of the cancer study, the validity of the instrument was determined by administering it to 10 articulate and educated family caregivers of the patients who were not involved in the main study. In completing this exercise, the content and face validity of this slightly modified version for relatives of SCD patients was tested by being presented for scrutiny to senior hematologists and specialty nurses at our hospital. They all approved the questionnaire. The burden instrument and the GHQ-12 were translated into the local Yoruba (also the language of the interviewer) by the method of back translation.

With experience of family caregiving roles in the locality,[18] there was no reason to suspect that relatives of cancer patients would be different from those of SCD patients in any way other than the illness variable. Hence, we judged that the questionnaire would be equally reliable in eliciting relevant responses from relatives of SCD patients. As active members of the culture, we have no reason to suspect that it is more socially acceptable to acknowledge financial burden than social or psychological burden. Previous Nigerian workers in the field had not noted it as an issue of concern.[13,26]

SUBJECTS AND PROCEDURE

The study involved interview of family caregivers who accompanied consecutive SCD patients to the adolescent/adult haematology clinic of the University College Hospital, Ibadan (UCH), Nigeria, for treatment. Relatives of patients in stable condition were interviewed at the clinic, while those of patients in sickle cell crisis were interviewed the next day at the nearby hematology Day Care Center, after completion of admission facilities. To be included in the study, the patient had to be accompanied to hospital by one or more adult family members (at least 16 years old) who were directly involved in informal caregiving roles at home (either living with the patient or seeing the patient regularly). In Nigeria, it is quite common for patients to be accompanied to hospital by relatives.[18] Also, as is usual in our culture for this type of study, all the patients and relatives approached, consented to be interviewed.

For each patient, the family member that was most intimately involved in caregiving roles was interviewed privately. Mothers of children were interviewed in cases where both parents brought the patient to the clinic. All interviews were conducted by a Nigerian senior female research nurse (on regular employment for that purpose), who also interviewed relatives for the earlier study of cancer patients. She was trained in the use of the study instrument by the senior author. Data collection commenced when we were satisfied that she had achieved high competence in reading out the items of the instrument in Yoruba (the local language), and rating the responses. In consideration of the relatively low literacy rate in our country, and to ensure uniformity, all the subjects had the items of the instrument read aloud to them, and the research nurse rated their responses. Subjects literate in English were interviewed in English, while the others were interviewed in the Yoruba language.

DATA ANALYSIS

Data were analyzed by SAS (Statistical Analysis System). Frequency distributions of sociodemographic characteristics and responses to burden questions were observed. We used parametric statistics because on inspection of frequency distribution, the data were fairly normally distributed. Caregivers of patients in SCD

crisis (as already defined) were then compared with those of patients in stable condition (i.e., not having been in crisis for at least six months), using *t*-tests for summary burden scores (to be defined later), age and GHQ scores; and Chi-square tests for socio-demographic characteristics. Statistical differences in summary burden scores and GHQ scores across marital status, level of education and occupational status were calculated using one-way analysis of variance (ANOVA), with Duncan's method of multiple comparison. Pearson's correlation was used to assess the relationship among burden indices, GHQ score, age of patients and caregivers.

Multiple regression analysis was used to assess factors that could predict global rating of burden, using the item on how much difficulty the relative experienced in coping with the patient's condition, as the dependent (or outcome) variable. In this regression analysis, the predictor (or independent) variables were: all summary burden scores, the GHQ score, the age of the patient and age of the caregiver. *T*-tests were two-tailed and the level of statistical significance was set at 0.05.

The summary burden score for each domain of objective burden (e.g., financial) was derived by adding up the scores on the relevant items. For each item, we had a 0–3 scoring system whereby "None at all"/"Not at all" = 0; while "occasionally so"/"mild difficulty" = 1, etc. As these domains did not contain an equal number of items, raw scores were measured by dividing the total scores by the number of items.

To rate clinical severity, we chose clinical items that have the potential to contribute directly to caregiver burden. For instance, in clinical practice, doctors give shorter appointment times to patients who are perceived to be still ill and require close attention. Also, the most common reason for hospitalizing patients is perceived severity of clinical condition; and in SCD, pain is the most common symptom. On the other hand, having to frequently bring patients to hospital, having them in hospital, and watching them in pain, are factors that have a direct impact on the caregivers' material and emotional resources. Hence, the severity of the patient's clinical condition, as an index of caregiver burden, was quantified by adding up the scores on frequency of hospital attendance, number of hospital admissions in the past year, and frequency of severe pain. This was operationally defined as "clinical severity burden." As a result, the following caregiver burden summary scores were obtained:

1. Clinical severity burden score: a measure of clinical severity of the illness.
2. Financial burden score: a measure of family's financial distress.
3. The weighted financial burden score was thus defined by dividing the financial burden score by five (i.e., number of items assessing financial burden).
4. The family routine burden score, a measure of disruption of family routines (e.g., difficulty in keeping to household work), was derived by adding up the scores on four related items.
5. The family disharmony score, a measure of the level of cordial relationships at home and with neighbors (e.g., quarrels, feelings of isolation by neighbors) was derived by adding up the score on three related items.
6. The weighted family burden score was derived by adding up the routine burden score and disharmony score, and dividing by seven.

Data analysis was guided by the following hypotheses: (1) caregivers of SCD patients in crisis will perceive significantly higher objective burden (e.g. financial and disruption of family routines) than caregivers of SCD patients in stable clinical condition; (2) financial burden is significantly higher than other burden indices; (3) majority of SCD caregivers will report low levels of subjective burden, with intact family emotional relationships and little or no social stigma attached to SCD; (4) SCD caregiver

global rating of burden (i.e. degree of difficulty of coping with SCD) is significantly correlated with objective burden indices and psychic distress; (5) compared with the results of a previous study in the same hospital, caregiver objective burden indices for SCD are lower than that for cancer, and SCD is associated with less social stigma.

RESULTS

Patients and caregivers of the two groups (i.e., in sickle cell crisis versus steady state) had similar socio-demographic characteristics and clinical severity indices (assessed in a longitudinal dimension).

Socio-Demographic Characteristics of SCD Patients and Caregivers (n = 81)

The majority of caregivers (58%) were mothers, and 74 (91.4%) lived in daily contact with the patients. There were no significant sex differences in age, socio-economic indices, indices of burden and GHQ-12 scores. The mean age of the patients who presented in sickle cell crisis (25.7, SD 10.6 years) was similar to those of patients in steady state (24.2, SD 12.8years) ($t = 0.5$, $p = 0.3$); and the mean ages of caregivers were also similar (40.4, SD 13.9years versus 41.1, SD 14.5 years) ($t = 0.2$, $p = 0.4$). In view of their age groups, the majority of patients were either students or junior civil servants, while the majority of caregivers had at least secondary school education (68.1.%) and were gainfully employed (59.3%) as civil servants, teachers, or in medium and large scale private businesses.

Severity of Burden (Tables 2 and 3)

Although the clinical severity scores were similar, relatives of patients in sickle cell crisis experienced significantly greater financial burden ($p < 0.02$) and family routine burden ($p < 0.05$). Interestingly, the GHQ scores were similar. When the results were combined, the weighted financial burden (1.8, SD 0.8) was significantly higher than the weighted family burden score (1.5, SD 0.5) ($t = 2.87$, $df = 160$, $p = 0.005$). It appears that this finding was largely contributed to by those in crisis, for whom the weighted financial burden (2.1, SD 0.7) was significantly higher than the weighted family burden score (1.7, SD 0.6) ($t = 2.1$, $df = 44$, $p = 0.04$). This indicates that the financial burden was much more problematic than the adverse effect of caring on family routines, especially for relatives of patients who presented with sickle cell crisis.

Apart from the primary effect on caregivers interviewed, other family members also had been similarly adversely affected in revenue generating activities (20 or 24.7%).

In spite of the objective burden that caregivers experienced, the vast majority admitted only a low level of subjective burden, as family relationships seemed to remain intact (88.9%) and there seemed to be little or no perceived social stigma attached to SCD (90.1%). On a

Table 1. Socio-demographic characteristics of all SCD patients and caregivers

A. Patients' characteristics	N = 81 (%)
1. Sex : male	44 (54.3%)
2. Occupation of patients : Employed	25 (30.8%)
-Students	38 (46.9%)
3. Age : (years): Mean (SD)	24.5 (12.1)
-Range: 9–20	36 (44.4%)
-21–30	33 (40.7%)
-31–40	5 (6.20%)
-41–85	7 (8.60%)
4. Level of education: No formal education	5 (6.2%)
5. Marital status: Single	66 (81.5%)
6. Relationship with caregiver: Mother	47 (58.0%)
-sibling	14 (17.3%)
-spouse/child	17 (20.9%)
7. Whom patient lives with: Parents	63 (77.8%)
-Sibling/Child	12 (14.8%)
B. Caregivers' characteristics	
8. Occupation: Unemployed/Petty trader	33 (40.7%)
9. Mean age (years) (SD)	41.1 (14.2)
10. Caregiver is married	58 (71.6%)
11. Caregiver lives with patient	74 (91.4%)

Table 2. Comparison of summary scores of indices of clinical severity and objective burden between caregivers of SCD in crisis and those not in crisis

Burden indices	Mean summary scores (SD)		Stats. differences		
	Not in crises (N = 57)	In crisis (N = 24)	t	df	P value
1. Clinical severity	4.4 (1.5)	4.5 (1.9)	0.2	79	0.8
2. Financial burden	8.2 (4.1)	10.6 (3.6)	2.4	79	0.02
3. Weighted financial burden	1.6 (0.8)	2.1 (0.7)	2.4	79	0.02
4. Family routine burden	6.8 (2.6)	8.2 (2.5)	2.0	79	0.05
5. Family disharmony score	3.4 (1.0)	3.7 (1.7)	0.8	79	0.4
6. Weighted family burden score	1.5 (0.6)	1.7 (0.6)	1.9	79	0.06
7. GHQ-12 score	1.3 (2.4)	1.2 (1.5)	0.3	79	0.8

NOTE: (a) For those in crises: weighted financial burden was significantly higher than weighted family burden score. (b) For the combined groups (i.e., crises + steady state) weighted financial burden was significantly higher than weighted family burden score ($t = 2.87$, $df = 160$, $p = 0.005$).

global rating of family difficulty with coping with SCD, the majority (47 or 57.9%) rated this difficulty as none or mild, while a quarter rated it as moderate.

Only 13 (16%) of the caregivers admitted that the caring had adverse effects on their physical health. The GHQ-12 symptom profile showed that eight (9.9%) caregivers admitted feeling unhappy and 11 (13.6%) felt stressed.

Factors Associated with Burden (Table 4)

Using the summary scores for Pearson's correlation analysis, we found that the degree of difficulty of coping with SCD (i.e., global rating of burden) was significantly correlated with financial distress, disruption of family routines ($p = 0.0001$) and age of caregiver ($p = 0.02$); but just failed to be significantly correlated with the psychological distress (GHQ)

Table 3. Frequency (%) of indices of clinical severity and psychosocial burden: all caregivers

Indices	Frequency (%), N = 81
1. Weekly/forthnightly hospital attendance	7 (8.6)
2. Not admitted in past one year	47 (58.0)
3. In severe pain most days of the week	10 (12.3)
4. Moderate/major loss of revenue by patient because ill	22 (27.2)
5. Moderate/major loss of revenue by family member on patient care	20 (24.7)
6. Expenditure incurred has moderate/major impact on family	29 (35.8)
7. Family taken moderate/major loan/sold property	14 (17.3)
8. Patient frequently/cannot attend work/school	18 (22.2)
9. Patient frequently has difficulty with household work	21 (25.9)
10. Patient's illness frequently disrupted caregiver's work	7 (8.6)
11. Frequently caring caused neglect of other family members	2 (2.5)
12. Caring never caused quarrel or disagreement at home	72 (88.9)
13. Family does not feel secluded/isolated/stigmatised	73 (90.1)
14. Global rating of family difficulty with coping	
-No difficulty	30 (37)
-Mild difficulty	17 (20.9)
-Moderate difficulty	21 (25.9)
-Severe difficulty	13 (16)

Table 4. Factors associated with overall feeling of burden by all caregivers

Factors/Variables	r	df	P value
A. Global rating of burden by caregiver			
-and family financial distress	0.54	71	0.0001
-and weighted family financial distress	0.54	71	0.0001
-and disruption of family routines	0.51	65	0.0001
-and family disharmony score	0.34	77	0.002
-and age of caregiver	0.28	70	0.02
-and age of patient	−0.15	78	NS
-and caregiver's GHQ-12 score	0.06	78	NS
B. Caregiver perception of clinical severity			
-and weighted family financial distress	0.46	55	0.0004
-and subjective burden of family	0.37	61	0.004
-and weighted subjective burden	0.37	61	0.004
-and GHQ-12 score of caregiver	0.24	69	0.052
C. Caregiver GHQ-12 score			
-and disruption of family routines	0.26	68	0.03

score (p = 0. 06). Caregiver perception of clinical severity of SCD just failed to be significantly correlated with psychological distress (p = 0.052), but was significantly correlated with family financial distress and subjective family burden. The factor that seemed to most adversely affect caregiver psychological distress score was disruption of family routines (p = 0.03) (see Table 4).

The results of one-way ANOVA operations showed notable significant differences in burden indices for caregiver levels of education and occupation. Although clinical severity scores showed no significant socio-demographic differences, caregivers with post-secondary levels of education experienced significantly lesser financial burden (f = 4.3 df = 2/78, p = 0.008). Those with higher employment status (civil servants and others) also experienced significantly lesser financial burden (f = 6.4, df = 2/78, p = 0.003) and lower GHQ scores (f = 5.7, df = 2/78, p = 0.005). The tendency for those with higher employment status (civil servants versus petty traders) to experience lesser family routine burden, did not reach significance (p = 0.08). The GHQ score of those caring for widows (5.0) was significantly higher than the score for those caring for patients who were married (1.4) or single(1.1) (f = 3.4, df = 2/78, p = 0.04).

In view of these multiple relationships, the data were subjected to multivariate statistics. In multiple regression analysis, global (overall) rating of burden by caregivers was entered as dependent variable, while the other factors were treated as independent variables. Global rating of burden was only significantly predicted by disruption of family routines (t = 9.6, b = 0.19, p = 0.003), and age of caregiver (t = 7.1, b = 0.02, p = 0.003), accounting for 28.9% of the variance.

Compared with the results of a similar study of relatives of patients with cancer at the same hospital, we found that, although the summary scores on disease clinical severity were much higher for cancer (7.6 versus 4.4), the two groups of caregivers had similar mean GHQ-12 scores (1.3 for cancer, and 1.3 for the current study). In addition, caregivers of SCD patients who presented in sickle cell crisis, and those for cancer, had very similar weighted financial burden scores (2.1 for SCD and 2.1 for cancer), and weighted family burden scores (1.7 for SCD and 1.8 for cancer). In comparing the emotional impact of the two diseases on the home environment, 72 (88.9%) and 70 (95.9%), respectively, of SCD and cancer caregivers, said that the fact of illness did not cause quarrels at home. Also, 73(90.1%) and 72(98.7%), respectively, of SCD and cancer

caregivers, did not feel that their families were socially stigmatized.

DISCUSSION

The findings should not be generalized, since the subjects were not a representative general population sample of SCD caregivers. However, the socio-demographic characteristics of SCD patients were similar to those of a previous study of adolescent/adult clinic attendees.[9] The caregivers interviewed had similar socio-demographic and psychopathological symptom (GHQ-12) scores as junior and middle level civil servants at Ibadan who were involved in another recent study.[27] Thus, there is no reason to suggest that the subjects involved in this study differed in any way from the general Nigerian urban population other than the illness variable. In psychosocial terms, the research evidence suggests that families with physically chronically-ill members do not significantly differ from those without illness.[28]

Another limitation of our study is the fact that only one caregiver was interviewed per family, so that it is reasonable to expect that a greater depth of information on family burden would be obtained by assessing multiple informants, including the patients' adaptation and opinion on burden. Like other workers,[13,17] we tried to overcome this problem by interviewing the family member most closely involved in the caregiving role. The inclusion of such a broad age range of patients is a limiting factor in this study, because developmental factors may require significantly different efforts by caregivers if the patient is a child, adolescent or adult. However, this is a naturalistic study in which we aimed to interview a variety of relatives of patients attending the clinic. In statistical analysis, the patient's age was not significantly correlated with burden indices. Rather, global rating of burden was significantly predicted by age of caregiver.

In pursuit of the first hypothesis, we found that although clinical severity scores and GHQ scores were similar between caregivers of SCD patients in crisis and those in stable condition, the former relatives experienced significantly higher scores on indices of objective burden, such as financial and family routine burdens. In relating this result with the fifth hypothesis, we found that caregivers of SCD patients in crises and those of cancer patients had very similar weighted financial burden and family burden scores. These results indicate that the objective burden of SCD in crisis is probably similar to that of cancer, and that what really makes SCD burdensome to relatives is the issue of frequent crises. It appears that if sickle cell crisis can be controlled, SCD may become a hidden disease in the social sense, just like properly controlled insulin-dependent diabetes mellitus.

In studies that have found higher levels of emotional disturbance among SCD patients, compared with diabetics,[29] it appears that what makes the difference is the painful crisis of SCD, the onset of which is preceded by severe depressive effects such as, helplessness and hopelessness.[30] One value of the comparison between caregivers of SCD in crises and those in stable condition, which clinicians would find interesting, is that it has enabled us to show the similarity of caregiver objective burden indices for SCD in crises and cancer, as well as the salutary effect of preventing SCD crises on caregiver burden in our locality.

With regard to the second hypothesis, we found that, in line with previous Nigerian studies on psychosocial burden,[19,26] the financial burden score was significantly higher than the other burden indices of family living. This is not surprising in the face of the inflationary trend in a severely depressed national economy with socio-political instability. The absence of national programs on medical insurance and social welfare makes this finding more worrisome. However, in analyzing for the fourth hypothesis, we found that, in multiple regression analysis, the financial burden score did not feature as a significant predictor of global rating of burden, a finding that is in marked contrast to the results for the cancer caregivers.

The significant predictors of burden in this

study were disruption of family routines and age of caregiver. This result is probably accounted for by the fact that, whereas for the cancer study, objective burden indices were similar across socio-demographic groups; in the case of SCD, higher occupational attainments significantly ameliorated financial burden. Hence, it could be argued that for the caregiver with higher occupational attainment, the most important factor was how to deploy resources to make sure that the family routines were stable, and to see that the impact of SCD on family routines did not adversely affect economic activities.

Other factors that moderated on perceived objective burden among our subjects were educational and occupational attainments. We found that, although there were no socio-demographic differences in clinical severity scores, relatives with higher educational and occupational attainments experienced significantly lesser financial and family burdens, as well as psychological distress. One possible implication of these findings is that the psychosocial burden of SCD can be significantly reduced by active attempts to control the frequency and duration of crisis, as well as providing adequate information and socio-economic supports to affected families.

With regard to the third hypothesis, we found that for the vast majority of caregivers, family relationships seemed to remain intact and there seemed to be no perceived social stigma attached to SCD. The context that enabled caregivers to report consistent dedication to their caregiving roles can be understood from the implications of the findings on coping strategies. There is an indication that, most of the time, relatives do not see problems as permanently insoluble.[31]

When compared with studies from the Western world, we found that the main difference with our findings is the social circumstance of caregivers. While the samples from the US and UK consisted predominantly of caregivers who were single mothers and others of relatively low social-economic status (SES),[14,17,32,33] our caregivers were predominantly married and their SES reflected the range of SES in Nigeria. However, the caregivers from the Western world had opportunity for medical insurance, social welfare services, strategically-located sickle cell centers and lay self-help groups.[5] These services are notably absent in Nigeria, except for a fledgling National Sickle Cell Association. The few sickle cell clubs pioneered by researchers are centered around teaching hospitals in large urban areas.[34]

Family life and employment are the most significant areas of social life in SCD, because financial status plays an important role in coping with physical disability.[15] Ievers[17] noted that both greater family cohesion and adaptability were found to buffer the potential negative effects of caring for SCD children with greater behavioral problems. Accordingly, it is reasonable to suggest that the fairly high level of family cohesion among our sample of caregivers (as evidenced by marital status and living arrangement data), coupled with the usual support of the extended family in our setting,[18] may have contributed to the finding that the majority (57.9%) rated their global family difficulty of coping with SCD, as "none" or "mild."

Like in all other studies of a similar nature, the majority of caregivers were mothers.[5,15] The assumption has been that the responsibility for the care of such children falls predominantly on mothers.[35] The results of a survey of the parents of children with SCD in Nigeria were broadly in line with this assumption; only 33% of fathers, compared with 80% of mothers, reported that their working lives had been affected by their child's condition.[6]

Our results indicate that, social support for caregivers, either in the form of financial assistance (e.g. national medical insurance), or stabilizing of family routines, early detection and prompt management of SCD crisis, and relief of psychological distress (e.g. social welfare programs and lay self-help group programs) is likely to relieve the psychosocial burden and make for better quality of care for SCD patients. At the moment, the only functional lay

self-help groups (e.g. National Sickle Cell Association) are based in a few teaching hospitals and are yet to have sufficient community impact. Our findings show that caregivers of patients in SCD crisis perceived similar financial and family routine burdens and psychological distress scores, in comparison with caregivers of cancer patients. A highlight was the possibility that active efforts to prevent SCD crises could lead to a stable condition of SCD where the burden of the disease could be similar to well-controlled diabetes mellitus.[29,32] One policy implication of these findings is that, preventive measures, such as genetic counseling and public health education on the risk factors for SCD crises should be promoted, while hematology staff should acquire competence in the early detection and management of the psychosocial complications of SCD.[36]

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