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Original Research Article

Psychosocial Burden of Childhood Sickle Cell Disease on Caregivers in Kenya

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Abstract

Objectives To characterize the types and magnitude of psychosocial burden present in caregivers who have a child with sickle cell disease (SCD) in Kenya and to identify predictors of caregiver psychosocial burden, including disease severity and financial hardship. **Methods** Primary caregivers ($N = 103$) of children aged 1–10 years diagnosed with SCD completed surveys assessing multiple domains of caregiver quality of life (QOL), adjustment to child illness, mental health, and financial hardship. Descriptive statistics characterize psychosocial burden, and linear models assess associations. **Results** On indicators of QOL, caregivers report multiple difficulties across most domains, including daily activities and physical, social, cognitive, and emotional well-being. Daily activities emerged as most burdensome. On indicators of parental adjustment to chronic illness, guilt and worry emerged as the greatest concern, followed by long-term uncertainty and unresolved sorrow and anger; relative to these, they reported higher levels of emotional resources. Financial hardship was high, as caregivers reported moderate to major financial losses due to the time spent caring for their child. General linear model analyses revealed that level of financial hardship was a significant predictor of all negative psychosocial outcomes. **Conclusions** Results document that Kenyan caregivers of children with SCD experience difficulties across multiple domains of functioning and that financial difficulties are likely associated with psychosocial burden. Results can guide intervention development for caregivers of children with SCD in low-resource, global contexts.

Key words: chronic illness; culture; disparities; family functioning; hematology; mental health; parent stress; parenting; psychosocial functioning; public health; quality of life; sickle cell disease; stress.

Introduction

Sickle cell disease (SCD) is one of the most common genetic disorders of the blood, producing abnormal hemoglobin molecules that result in rigid, sticky, sickle-shaped cells (Edwards et al., 2005). SCD is a chronic condition that can cause painful episodes, termed pain crises, and progressive damage to multiple organ systems (Rees, Williams, & Gladwin, 2010). Pain crises can be severe, are usually impossible to anticipate, and vary in duration, location, type, and

severity (Fuggle, Shand, Gill, & Davies, 1996). The 63rd meeting of the UN General Assembly in 2008 accepted a declaration on the "recognition of sickle-cell anaemia as a public health problem" and urged other Member States to raise awareness of SCD (Grosse et al., 2011). Although this was a major step in recognizing the global impact of the disease, relatively little attention has been given to assessing the psychosocial burden on children and families outside of high-income countries.

Globally, over 300,000 children are born with SCD each year, and approximately 70% are in Africa where very little research has been conducted (Grosse et al., 2011; Modell & Darlison, 2008). In many low- and middle-income countries (LMICs), including Kenya, healthcare resources and methods for early detection are limited, leading to undiagnosed and untreated SCD that often results in early childhood mortality (Piel et al., 2013). In a 2013 study, it was estimated that 50–90% of infants born with SCD in Africa died before age 5 (McGann et al., 2013). There have, however, been some encouraging shifts, including the increasing use of hydroxyurea across Africa, including in Kenya (Tshilolo et al., 2019). Such advances have led to increased life expectancy for patients with SCD, with some surviving until reproductive age (Tshilolo et al., 2019). With these achievements, it is now especially clear that attention is needed towards better understanding the psychosocial dimensions of the illness to promote quality of life (QOL) for children living with SCD in LMICs and their families (Ohaeri & Shokunbi, 2002). The field of pediatric psychology is well-positioned to lead this effort.

SCD can place significant psychosocial burden on children and their families across settings (Anie, 2005; Barakat, Lash, Lutz, & Nicolaou, 2006). The term "psychosocial burden" in this context refers to a broad range of stressors and associated psychological distress, behavioral difficulties, and problems related to relationships and social functioning (Barakat et al., 2006). For children, challenges vary across developmental stages and include reduced participation in recreational activities, high levels of school absenteeism, delayed puberty, limitations in social functioning, and low overall QOL (Hildenbrand, Barakat, Alderfer, & Marsac, 2015). In addition to child-level stressors, caring for a child with a chronic illness can have psychosocial consequences through innumerable stressors—the overwhelming news of diagnosis, medical risks and costs, time required for care and appointments, and, at times, shortened life expectancy (Cousino & Hazen, 2013). Not surprisingly, parents of youth with SCD experience psychological symptoms, including symptoms of depression and anxiety (Moskowitz et al., 2007). As an example, female caregivers in the Netherlands exhibited low Health-Related Quality of Life (HRQOL) characterized by depressive moods, disturbances in daily activities, vitality, sleep, happiness, and cognitive functioning (van den Tweel et al., 2008). Among families of children with SCD in Saudi Arabia, Madani, Al Raddadi, Al Jaouni, Omer, and Al Awa (2018) also found high rates of exhaustion and unmet financial needs. In the USA, greater symptoms of distress in a caregiver has been linked to diminished QOL for the child (Panepinto, Hoffmann, & Pajewski, 2010).

There is a paucity of research examining the experiences of caregivers of children with SCD in LMICs, particularly in Africa where the disease is most prevalent. The small number of studies available do show evidence of psychosocial burden. One study in Nigeria examined the family-related psychosocial burden of SCD, positing that the impact on family is worse in LMICs due to inadequate social welfare and healthcare services. Within a sample of 225 caregivers, they found that family members of children and adolescents with SCD experienced significant financial stress, depressive symptoms, and neglect from other extended family members (Adegoke & Kuteyi, 2012). In Cameroon, parents of children and adolescents with SCD ($N = 130$) experienced moderate to severe difficulties in coping, with clinical severity emerging as a salient factor influencing ability to cope (Wonkam et al., 2014). Lastly, in Kenya, a qualitative study with 13 families documented stigmatization surrounding SCD resulting in negative consequences, especially for mothers; participants reported mothers are often blamed for the disease and endure effects on their livelihoods due to the interactions between poverty and demands of seeking care (Marsh, Kamuya, & Molyneux, 2011).

The current study takes a next step in this research by examining a broader range of psychosocial sequelae of caring for a child with SCD in a LMIC setting and by beginning to examine predictors of this burden. We first aim to characterize the types and magnitude of psychosocial burden experienced by caregivers of children aged 1–10 across multiple domains related to physical, social, and emotional well-being. To do this, we explore measures of QOL and parental adjustment to children's illness. We also describe financial hardship associated with caregiving demands. We then examine potential predictors of caregiver psychosocial burden, including financial hardship, disease severity indicators (i.e., pain crises and hospitalizations), and the age and gender of the child. Based on previous literature, we hypothesized that caregivers in this setting would experience psychosocial burden across domains and that higher levels of financial hardship and more severe disease would be associated with higher psychosocial burden. Results begin to provide insight for the development

of interventions to reduce the impact of SCD on families in Kenya and other LMIC contexts.

Methods

Setting

The study took place in Homa Bay, Kenya at Homa Bay County Teaching and Referral Hospital. Homa Bay is in Western Kenya on the border of Lake Victoria, with a county poverty rate estimated at 44% in 2013(KNBS and UNICEF, 2013). This location is holoendemic for *Plasmodium falciparum* malaria, with an estimated prevalence rate of over 30% among children aged 2–10 years (Macharia et al., 2018). The prevalence of the HbS allele is also high, estimated to be above 15% in the areas surrounding Lake Victoria—significantly higher than the 7–12.5% prevalence in other areas of Sub-Saharan Africa (Piel et al., 2013). The combination of these diseases produces compounded health risks for children, and malaria often increases sickle cell pain (Fleming, 1989). The Sickle Cell Clinic in Homa Bay operates through a collaboration between Academic Model Providing Access to Health Care (AMPATH) HematoOncology, Moi Teaching and Referral Hospital (MTRH) in Eldoret, Kenya, and the Homa Bay County Government.

Participants and Procedures

Participants were recruited whose children were enrolled in a clinical trial (Enhancing Preventative Therapy of Malaria in children with Sickle cell anemia in East Africa [EPiTOMISE], NCT03178643) that aims to compare the efficacy of medications to prevent *P. falciparum* malaria in children with SCD. Any caregiver of a child enrolled in EPiTOMISE was eligible to participate in this study. In very rare cases of two caregivers attending, the one who provided child permission for EPiTOMISE was included. Inclusion criteria for the trial for children included: age between 12 months and 10 years; residence in malaria-endemic counties of Homa Bay or Migori, a neighboring location; hemoglobin SS confirmed by electrophoresis; and caregiver consent. Exclusion criteria included: taking routine antimalarial prophylaxis for an indication other than SCD; other chronic conditions; and/or blood transfusion in the prior 120 days. In cases of recent transfusion, the child could then be enrolled after 120 days had passed. During their 12-month participation in the trial, children visited the study clinic monthly and received free study medications and enrollment in the Kenya National Hospital Insurance Fund. As participants reported for clinical trial visits, caregivers were invited to participate in this substudy by research assistants (RAs) fluent in English, Kiswahili, and Dholuo, the local language. After obtaining signed informed consent, interviews were conducted in the language most comfortable for the participant. All procedures were approved by Duke University and Moi University College of Health Sciences institutional review boards.

Sample Size Calculation

Sample size was calculated using the power function in R (Champely et al., 2018). Eighty participants were needed to achieve 80% power to detect a minimum effect size of 0.15 at an alpha level of 0.05 for linear models. The final sample included 103 caregivers, increasing power to 90% to detect this same effect size.

Measures

Adaptation and Development

As most measures were developed in the US, we translated, piloted, and made necessary adaptations to all measures prior to data collection. Items were first translated from English into Kiswahili and Dholuo by RAs fluent in all three languages. They were then back translated to check for loss of meaning or problems in wording. We then conducted interviews with 11 pairs of caregivers ($N = 22$) to assess items for comprehensibility, acceptability, and relevance in this context. Guided by van Ommeren's technique for transcultural translations (van Ommeren et al., 1999), RAs presented items one-by-one and asked the pair whether it was understandable, whether they would feel comfortable answering, and whether they believed it to be relevant to the local context. Each pair reviewed approximately 15 items. If an item did not meet all three criteria, participants were asked to suggest changes. It was then revised and presented to subsequent pairs until it passed all criteria. For example, the wording of the item "I feel sick to my stomach" was difficult to understand. Participants suggested "I feel pain in my stomach," which passed the criteria with the next pair. Using this process, the wording was revised for nine items, including six from the Pediatric Quality of Life Family Impact Module (Peds-QL FIM; one Physical, three Emotional, one Social, and one Cognitive), one item on the Parent Experience of Child's Illness Scale (PECI) on the Unresolved Sorrow and Anger subscale, one financial hardship item, and one Patient Health Questionnaire-2 (PHQ-2) item.

In addition to testing the standardized items, we conducted a free listing exercise with the same participants, except one who needed to leave early, to determine if new items should be added for this context. Participants were first asked to list anything that was not covered in the items they reviewed that caused them stress and/or hardship. Second, they were asked to list anything positive related to caring for a child with SCD to assess for positive or growth-oriented aspects of the caregiving experience. The lead researcher reviewed the lists, grouping together

responses that were similar in content. If three people mentioned similar topics, a new item related to that construct was developed and added as a "locally-developed item." As a result of this process, nine new "local" items were added (described below).

The Pediatric Quality of Life Family Impact Module

The Peds-QL FIM measures the impact of pediatric chronic health conditions on parents and families (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004). This 36-item questionnaire has six subscales measuring caregiver self-reported functioning across multiple domains: Physical (6 items; Chronbach's alpha (α) = .86 in this sample), Emotional (5 items; α = .71), Social (4 items; α = .71), Cognitive (5 items; α = .81), Communication (3 items; α = .39), Worry (5 items; α = .69), Daily Activities (3 items; α = .85), and Family Relationships (5 items; α = .75). A 5-point Likert-type scale is used from 0, "Never a problem," to 4, "Almost Always." Items are reverse-scored and linearly transformed to a 0–100 scale such that higher scores indicate better functioning. The Peds-QL FIM Total Score is the sum of all 36 items divided by the total number of items answered (α = .84). The parent HRQOL Summary Score (20 items) is computed as the sum of the items divided by the number of items answered in the Physical, Emotional, Social, and Cognitive Functioning subscales (α = .88). There also is a Family Functioning Score (eight items) generated from Daily Activities and Family Relationships items, but we did not use this due to low internal consistency (α = .21).

The Parent Experience of Child's Illness Scale

The PECI is a 25-item self-report measure of a parent's adjustment to their child's chronic illness (Bonner et al., 2006). It includes 25 items and 4 subscales, with some items loading onto multiple subscales. Subscales include Guilt and Worry (11 items; α = .69 in this sample), Unresolved Sorrow and Anger (8 items; α = .52), Long-Term Uncertainty (5 items; α = .58), and Emotional Resources (5 items; α = .41). Items are rated on a 5-point Likert-type scale from 0, "Never," to 4, "Always." Subscale scores are calculated by computing the mean.

Financial Hardship Assessment

To assess impact on family finances, a set of questions was adapted from a study on psychosocial burden of SCD on the family in Nigeria (Adegoke & Kuteyi, 2012). Four items related to the extent to which their child's SCD has impacted their finances, including: whether the family lost revenue, whether time spent caring for the child impacted financial benefits, whether the money spent on medical care impacted the family, and whether the family had taken out loans or sold valuable property to meet expenses related to the child's illness. Items asking about loss in revenue and impacted financial benefits were rated as: 0, "No Loss"; 1, "Minor Loss"; 2, "Moderate Loss"; and 3, "Major Loss." Choices for the item about the general impact on family were: 0, "None;" 1, "No, has affected but not significantly;" 2, "Has affected but we can cope;" and 3, "Has affected and we cannot cope." Lastly, choices for the item regarding loans were: 0, "Not at all;" 1, "Only small amounts;" 2, "Moderately large sum which the family is having difficulties paying back;" and 3, "A large sum of money which the family may not be able to repay." An overall financial hardship assessment score was calculated by computing the mean, with higher scores indicating higher burden. Families were also asked who is primarily responsible for settling the hospital bills.

The Patient Health Questionnaire-2

The PHQ-2 includes two questions about the frequency of depressed mood and anhedonia over the previous 2 weeks (r = .65 in this sample) (Kroenke, Spitzer, & Williams, 2003). Respondents use a 4-point scale: 0, "Not at all"; 1, "Several days"; 2, "More than half the days"; 3, "Nearly every day." The total score ranges from 0 to 6, with scores of 3 or above indicating likely symptoms of depression according to U.S.-based norms. The PHQ-2 has been used and validated in a Kenyan sample (Monahan et al., 2009).

Locally Developed Items

The nine new items added to the questionnaire included four related to burden and five to positive experiences, all structured with 4-point Likert scale response choices that mirrored the structure of the PECI. Burden items related to medication access, transportation access, stigma about community perception, and myths about origins of the disease; higher ratings indicated more burden. Positive experiences related to raising awareness about SCD, satisfaction with medications and treatment, knowledge about SCD helping other children or family members (e.g., encouraging others be tested), confidence in their child's survival, and future plans; higher scores indicated more positive experiences. Responses were analyzed only as single items for aim 1—descriptive analyses—and were not included in general linear models.

Demographics and Disease Severity Indicators

Demographic data for caregivers and children, as well as indicators related to disease severity, were collected as part of the overarching clinical trial through caregiver self-report. Items were verbally elicited from caregivers, and a member of the study team recorded the answers. Demographic information included: child age, child sex, child education status, caregiver

employment, caregiver education status, and family history of SCD. Caregiver-reported indicators of disease severity included number of pain crises and number of hospitalizations in the past 12 months, as well as number of school absences within the previous 30-day period due to a medical problem.

Data Analysis

Data were analyzed using R version 3.5.1 (R Core Team, 2018). Descriptive statistics were calculated to characterize the levels of caregiver psychosocial burden on all subscales and composite scores on the measures described above. General linear modeling was used to determine whether child age, child gender, number of pain crises, and financial hardship predicted caregiver psychosocial burden across domains. For linear model analyses, we focused on the subscales of each measure rather than the composites. The Communication subscale of the Peds-QL FIM and the Emotional Resources subscale of the PECI were not included in the linear model analyses given low internal consistency. There were no missing data.

Results

Participant Characteristics

Participants were 103 caregivers of pediatric SCD patients, which included those involved in measures adaptation. Characteristics are reported in Table I. The majority were mothers (72.65%). Most caregivers had at least a primary school education, with about half having post-primary schooling; half worked outside the home (50.49%). The average child age was 5.39 years, and all except one of the children of typical school age (over age 5) were enrolled in school. Among the 72 in school, they missed an average of 4 days of school in the past 30 days due to a medical problem. Nearly 43% had been hospitalized at least once in the past year for SCD complications, and almost 60% reported at least 3 pain crises in the past year; 8.7% reported more than 10. Approximately 60% were using hydroxyurea. Twenty had a sibling with SCD, nine of whom were deceased.

Table I. *Demographic Characteristics*

	M	*SD*
Age of child ($n = 103$)	5.39	2.61
Child sex	*N*	%
Female	39	37.9
Child education levels		
Not in school[a]	31	30.1
Nursery	40	38.8
Class 1	13	12.6
Class 2	10	9.7
Class 3	6	5.8
Class 4	1	1.0
Class 5	2	1.9
Number pain crises[b] (past 12 months)		
0 times	17	16.5
1 time	13	12.6
2 times	12	11.7
3–5 times	43	41.8
6–10 times	9	8.7
More than 10 times	9	8.7
Caregiver relationship to child		
Mother	84	81.6
Father	17	16.5
Aunty	1	1.0
Grandparent	1	1.0
Caregiver highest education completed		
No education	2	1.9
Primary	44	42.7
Some secondary	15	14.5
Secondary completed	23	22.3
College	20	19.4
Caregiver employment status		
Outside the home	52	50.5

[a]All except one of the children not in school were under the age of 5 (below the typical age children begin primary school).

[b]Pain crisis is defined as pain lasting 2 hr or more that did not have an obvious cause.

Aim 1: Types and Magnitude of Caregiver Burden

Domains of caregiver psychosocial burden are described here with full results in Table II.

Quality of Life

Participants reported difficulties across the majority of QOL domains as reflected in the composite and subscale scores on the Peds-QL FIM. The range is 0–100, with higher scores indicating better QOL, and the majority of scores for this sample fell below 50 (Mean [*M*] Total Score = 43.76). Participants reported the lowest QOL on the Daily Activities subscale ($M = 25.00$), in which approximately 70% reported "Almost Always" having difficulty with family activities taking more time and effort, finding time to finish household tasks, and feeling too tired to finish chores around the house. Participants' scores also reflected multiple difficulties on the Physical, Emotional, Social, and Cognitive QOL domains. We examined some single items to identify specific problems that were heavily endorsed by the participants. For example, on the Physical subscale ($M = 36.49$), 41% reported "Almost Always" feeling tired during the day. On the Emotional subscale ($M = 40.44$), the majority (58%) reported "Almost Always" feeling anxious. For Social Functioning ($M = 36.65$), the majority (52%) endorsed "Almost Always" finding it difficult to schedule time for social activities. Lastly, on the Cognitive subscale ($M = 42.77$), 45% reported "Almost Always" finding it hard to keep their attention on daily activities. Caregivers reported fewer concerns related to items on Family Relationships and Communication. On Family Relationships ($M = 58.50$), participants reported that they did not

Table II. *Mean and Standard Deviation Scores of Standardized Surveys*

Scales	*M* (*SD*)
Peds-QL FIM (range 0–100) (+)	
Physical	36.49 (27.76)
Emotional	40.44 (23.79)
Social	36.65 (25.99)
Cognitive	42.77 (27.26)
Communication[a]	62.30 (22.92)
Worry	47.91 (23.97)
Daily activities	25.00 (25.99)
Family relationships	58.50 (25.17)
Parent HRQOL score[b]	39.09 (22.37)
Total impact score[c]	43.76 (17.53)
PECI (range 0–4)	
Guilt/worry (−)	2.23 (0.65)
Long-term uncertainty (−)	1.75 (0.66)
Unresolved Sor/anger (−)	1.97 (0.86)
Emotional resources (+)[a]	2.59 (0.72)
Financial hardship (range 0–4) (−)	
Lost revenue	2.31 (0.84)
Loss due to time spent caring for child	2.33 (0.89)
Money spent due to illness affected family	1.76 (0.65)
Taken out loans or sold property to meet expenditure	1.66 (0.91)
Total score	2.01 (0.63)
Local burden indicators (range 0–4) (−)	
Community perception	1.82 (1.22)
Access to resources	1.49 (1.03)
Local positive experience indicators (range 0–4) (+)	
Access to medication	3.60 (0.82)
Knowledge about SCD has helped care for other children	3.57 (0.92)
Create awareness about the disease	3.70 (0.64)
Happy that child has survived	3.53 (0.97)
Confident that child will have a bright future	3.62 (0.82)
PHQ-2 (range 0–6) (−)	
Little interest/pleasure in doing things	1.79 (1.07)
Feeling down, depressed, or hopeless	1.94 (1.09)
Total score	3.74 (1.97)

Note. HRQOL = Health-Related Quality of Life; PECI = Parent Experience of Child's Illness Scale; PedsQL FIM = Pediatric Quality of Life Family Impact Module; PHQ-2 = Patient Health Questionnaire-2; SCD = sickle cell disease. (+) indicates higher scores corresponding to better outcomes; (−) indicates higher scores indicate worse outcomes.

[a]Subscale score had internal consistency values of 0.4 or below.

[b]HRQOL is the average of the 20 items on the Physical, Emotional, Social, and Cognitive Functioning subscales.

[c]Total score is the average of all 36 items.

often experience conflicts between family members. On Communication items, participants also reported few difficulties communicating with doctors and nurses or talking about their child's health with others in the community, though the overall scale had low internal consistency reliability.

On locally developed items, the majority reported infrequently experiencing shame or others avoiding them due to the illness, though this still occurred for about one-quarter of the sample; 54% said this never happens, 22% said "rarely" or "sometimes," and 25% said "often" or "always." Likely related, 38% of participants reported that other people "always" blamed SCD on witchcraft or evil spirits instead of knowing the origins of the disease, and an additional 34% responded "sometimes" or "often" to this item.

Parent Adjustment Related to Child Illness

On the PECI, participants' highest scores were on the Guilt and Worry subscale ($M = 2.23$). In particular, 55% of caregivers endorsed "always" worrying that something bad will happen to their child when they are out of their care. In addition, 82% reported "often" or "always" waking up during the night to check on their children. Long-term Uncertainty ($M = 1.75$) and Unresolved Sorrow and Anger ($M = 1.97$) scores also indicate that caregivers frequently experience difficulty within these domains. On Long-term Uncertainty, 80% of caregivers endorsed "often" or "always" worrying about whether their child will be able to live independently as an adult. Responses on the Emotional Resources items showed a somewhat more positive result, though low internal consistency reliability pointed us toward

examination of specific items rather than the overall score. Most caregivers reported that they were "always" ready to face challenges related to their child's well-being (64%), and 40% reported that they can "often" or "always" get help when and support when needed.

Financial Hardship

Caregivers were most impacted by losing revenue due to factors related to their child's illness through losing a job, losing working hours for business, or missing opportunities to go to the farm or market to sell; over half (52%) reported a "Major Loss" of revenue and just over one-quarter reported "Moderate Loss." Similarly, most caregivers reported losing general financial resources due to time spent caring for the child with SCD; again 57% reported "Major Loss," and roughly a quarter—22%—reported "Moderate Loss." When asked generally about financial impact and coping, nearly 80% chose the option stating they are "affected but are able to cope." When asked whether they had to take out loans or sell property to cover medical expenses, 55% reported receiving loans, which the family is having difficulties paying back, or selling property worth "moderately large" sums. A smaller but notable number (14%) reported owing "large sums which they may not be able to repay." When asked who settles the hospital bills for the child, 89% indicated the mother or father. For locally developed indicators related to resources access, 59% reported never worrying about transportation to receive care during a pain crisis, with 41% reporting some level of difficulty.

Locally Developed Positive Experience Indicators

Positive indicators were endorsed at a high rate. For all five items, at least 75% of caregivers reported "always" experiencing these positive indicators related to caring for a child with SCD, including feeling confident about their child's survival and future plans, better equipped to care for other children and family members, happy about creating awareness about the disease, and satisfied with access to medications (though this last indicator was likely a direct result of being part of the larger clinical trial).

Depression

The mean PHQ-2 score was 3.74 with the majority (69.53%) scoring a 3 or above, indicating likely symptoms of depression based on U.S. norms that were also applied in previous research in Kenya (Kroenke et al., 2003; Monahan et al., 2009).

Aim 2: Predictors of Caregiver Psychosocial Burden

General linear models were generated with each of the burden indicators as the outcome and child age, child gender, financial hardship score, and number of pain crises as independent variables. See Table III for full results.

Quality of Life

Higher financial hardship was associated with poorer QOL for all subdomains. Higher number of pain crises also emerged as a significant predictor of poorer Physical QOL. Pain crises were not associated with other subscale scores at an alpha level of 0.05, though the association was in the expected direction, or very near zero, for all except Family Relationships. Child age was associated only with Daily Activities, indicating potentially more difficulties for caregivers of older children. Child gender was not associated with QOL scores.

Parental Adjustment

Financial hardship was a significant predictor of higher scores on all subscales included in the analyses: Guilt and Worry, Unresolved Sorrow and Anger, and Long-term Uncertainty. Pain crises and child gender were not associated significantly with parental adjustment.

Caregiver Mental Health

Financial hardship was associated significantly with scores on the PHQ-2 assessing depressive symptoms, with higher hardship associated with higher symptom scores.

Discussion

This study examined caregiving stress and burden associated with pediatric SCD in Kenya. Results add to the small number of studies characterizing the types and magnitude of caregiver psychosocial burden in LMICs, Africa specifically, where SCD is most prevalent. Findings reflect that caregivers in this setting are indeed experiencing significant stressors and difficulties across multiple domains of QOL. These caregivers reported the greatest difficulty with practical struggles in daily activities, and endorsed items associated with high levels of worry about their children. Caregivers also reported significant financial hardship related to having a child with SCD, most often reported as losing revenue due to job loss and working fewer hours. Importantly, many caregivers also reported experiencing symptoms of depression. Alongside these difficulties, caregivers did indicate some strengths and positive experiences, including not feeling stigmatized in the community, feeling at peace with their

Table III. *General Linear Models: Predictors of Caregiver Psychosocial Burden*

Variable	Adjusted R^2	df	*F*-statistic	Estimate	*P*-value
Peds-QL FIM					
Physical score	0.21	4, 98	7.80		
Child age				0.71	0.45
Child gender				0.65	0.90
Financial hardship				−19.84	2.55E−06*
Number of pain crises				−3.26	0.05*
Emotional score	0.30	4, 98	11.78		
Child age				1.20	0.12
Child gender				3.21	0.43
Financial hardship				−19.98	9.35E−09*
Number of pain crises				−1.92	0.15
Social score	0.31	4, 98	12.58		
Child age				1.03	0.2
Child gender				0.03	0.995
Financial hardship				−23.57	8.56E−10*
Number of pain crises				−1.72	0.24
Cognitive score	0.13	4, 98	4.71		
Child age				0.58	0.55
Child gender				−2.01	0.70
Financial hardship				−17.29	5.54E−05*
Number of pain crises				−1.23	0.47
Worry score	0.07	4, 98	2.90		
Child age				0.33	0.71
Child gender				−0.86	0.86
Financial hardship				−12.26	0.001*
Number of pain crises				−0.87	0.58
Daily activities score	0.28	4, 98	10.98		
Child age				2.05	0.02*
Child gender				1.11	0.81
Financial hardship				−21.82	1.67E−08*
Number of pain crises				−1.17	0.43
Family relationships score	0.04	4, 98	2.09		
Child age				0.59	0.54
Child gender				−8.41	0.10
Financial hardship				−10.19	0.01*
Number of pain crises				0.96	0.56
PECI					
Guilt and Worry	0.18	4, 98	7.876.658		
Child age				−0.04	0.11
Child gender				−0.14	0.25
Financial hardship				0.43	1.47E−05*
Number of pain crises				0.01	0.71
Unresolved sorrow/anger	0.06	4, 98	2.77		
Child age				−0.02	0.43
Child gender				−0.02	0.90
Financial hardship				0.31	0.003*
Number of pain crises				0.04	0.37
Long-term uncertainty	0.06	4, 98	2.76		
Child age				−0.02	0.44
Child gender				0.34	0.05*
Financial hardship				0.37	0.01*
Number of pain crises				0.05	0.37
Depression symptoms					
PHQ-2 score	0.21	4, 97	7.68		
Child age				−0.03	0.63
Child gender				0.11	0.77
Financial hardship				1.55	3.05E−07*
Number of pain crises				−0.05	0.64

Note. df = degrees of freedom; PECI = Parent Experience of Child's Illness Scale; PedsQL FIM = Pediatric Quality of Life Family Impact Module; PHQ-2 = Patient Health Questionnaire-2.

*$p < 0.05$ (no adjustment for multiple comparisons).

circumstances, and having strong communication and family relationships. Many caregivers also reported confidence about their child's future despite the high levels of worry reported in this sample; this should be explored further, as caregivers may have meaningful ways of maintaining optimism and hope alongside their distress.

Compared with results of other studies in LMIC settings, these findings were largely consistent in documenting multi-faceted difficulties faced by caregivers of youth with SCD, including effects on economic well-being (Adegoke & Kuteyi, 2012; Marsh et al., 2011). One particular area of difference, however, was our finding that caregivers reported low levels of perceived stigma related to their child's SCD despite high endorsement of SCD being caused by witchcraft or evil spirits. In contrast, a qualitative study by Marsh et al. (2011) documented some families experience stigma, including parental blame for the disease, that has meaningful effects—in particular for mothers. This is an area for further investigation given its important implications for interventions at the structural, financial, and psychosocial levels.

Results also serve to document that caregivers of children with SCD in Kenya share many of the same stressors and threats to QOL that affect caregivers in high-income countries, though there may also be some differences to explore in future work. For instance, compared with three samples of caregivers of children and adolescents with SCD in the US (Allen, Anderson, Brotkin, Rothman, & Bonner, 2019; Anderson et al., 2018; Sabhlok, Anderson, & Bonner, 2016), this Kenyan sample endorsed greater burden on the Guilt/Worry subscale of the PECI, indicating more acute concern and anxiety related to their child's disease. Related to QOL, Panepinto, Hoffmann, and Pajewski (2009) found that a U.S.-based sample of caregivers reported relatively high burden on the Worry and Communication domains. Worry items also emerged as a concern in this Kenyan sample, but Kenyan caregivers reported few difficulties on Communication related to family and provider communication. These types of differences—though not precise given differences in the samples—are likely due to a myriad of factors, including cultural and contextual considerations, which should be a focus of future research to better inform transferability of interventions.

Our second aim was to conduct a preliminary examination of some factors that could predict psychosocial burden experienced by these caregivers. Findings showed associations between financial hardship and all parental adjustment and QOL domains. While this was not unexpected, it was somewhat remarkable in the consistency of results across the domains. As in our study, previous literature in LMICs has documented the clear economic impacts of both acute and chronic health conditions on households (McIntyre, Thiede, Dahlgren, & Whitehead, 2006; Okediji et al., 2017). Families, especially mothers, have also reported relatively few options in maintaining income generating activities, exacerbating financial and psychosocial problems (Marsh et al., 2011). These risks can lead to significantly lower HRQOL and increase psychological distress (Thrush & Hyder, 2014). A study conducted in the US further showed that individual socioeconomic status and neighborhood economic distress both predicted poorer functioning among children with SCD (Palermo, Riley, & Mitchell, 2008). Importantly, results across studies reflect that financial hardships go beyond the direct effects of medical care costs themselves, greatly impacting caregivers' income generating activities. Future research should more closely examine these experiences to identify potential points of intervention to reduce burdens related to both time and finances that are feasible in LMIC contexts.

More than half of the sample reported symptoms indicating possible depression, presenting a major cause for concern. Along with significant difficulties in QOL, this points to the potential utility of examining cycles of negative cognitions, emotions, and behaviors that could influence coping behaviors. For instance, a U.S.-based study by Sil, Dampier, and Cohen (2016) discusses pain catastrophizing—an exaggerated negative "mental set" in response to pain—as a maladaptive thought pattern that may result in increased emotional distress for the parent (Sullivan et al., 2001). Examining whether this or other patterns are also present in a Kenyan sample, or among others across contexts, would be valuable for informing treatments.

There are several limitations of this study to consider. First, we had no comparison group to compare QOL between caregivers of children with SCD and without SCD in this setting. Nor did we have a group of caregivers of children with other health conditions. These types of comparisons could help differentiate between unique burden associated with SCD versus psychosocial burden that generalizes more broadly and could be related to overall stressors, such as poverty, common in LMIC settings. There were also limitations of the sample, including a restricted age range that did not include caregivers of adolescents who face unique challenges associated with both developmental stage and the transition from pediatric to adult care (Poku, Caress, & Kirk, 2018). This is an especially interesting group for future research in a context where survival into adolescence still remains less common than in high-income countries (Grosse et al., 2011). In addition, the sample was drawn from one relatively rural location, the majority of children were male, and children received more consistent medical care as part

of the overarching study than other families might outside of the study context. Lastly, while the use of standardized tools with a systematic adaptation process is a strength of this study, some subscales demonstrated low internal consistency, pointing to the need for further measures development, adaptation, and validation. Mixed-methods studies with a qualitative component would have dual benefits of better understanding caregiver experiences and identifying context-specific indicators of psychosocial burden to improve measures.

While recognizing the limitations, findings are useful for informing the process of developing ideas and hypotheses for pediatric psychology intervention research for this population. Here, we present a set of ideas with particular focus on approaches likely to be feasible first steps for work in LMICs, recognizing the potential for the transfer of some intervention strategies from HIC while also considering the scarcity of mental health professionals. As a first step, to respond to overall QOL and adjustment concerns, interventions could focus on developing methods for healthcare providers to acknowledge and normalize caregiver distress, provide basic psychoeducation, and to facilitate problem-solving for some stressors caregivers are experiencing (e.g., identifying ways to minimize disruption to employment due to medical appointments). Alongside this, peer support interventions may help reduce stress and build coping strategies. Support groups for caregivers of children with chronic illness have been shown to foster a shared social identity, resulting in a sense of belonging, support, and empowerment, as well as improvements in family functioning and psychological health (Shilling et al., 2013). Near the site of this study, some caregivers have started such groups, showing the potential acceptability of more structured psychosocial interventions.

To address both illness-related stressors and depression symptoms, identifying ways to improve mental health assessment and treatment services is essential. For treatment, problem-solving therapy (PST) and cognitive behavioral therapy (CBT) are evidence-based options to consider. PST has been shown to promote positive mental health in caregivers of youth with chronic illness in the USA, including those with chronic pain (Palermo et al., 2016), and CBT is a well-established depression treatment (Cuijpers et al., 2013). Both have been shown to be feasible and effective in LMICs primarily through task-shifting approaches to nonprofessionals—approaches that hold promise for family-based approaches as well (Healy, Kaiser, & Puffer, 2018; Singla et al., 2017). In addition, given the influence of financial hardship, combination interventions that include both psychosocial and economic strengthening components should be considered.

Across intervention possibilities, attention is needed towards building training resources for pediatric psychology in LMICs to allow for active collaborations and scale-up. Furthermore, to facilitate dissemination and scale-up, pediatric psychologists should consider technology as a means to improve access and efficiency, especially given the proliferation of mobile phones and promising use of mobile technology for psychological interventions in LMICs (Bockting, Williams, Carswell, & Grech, 2016). Several U.S.-based studies have documented the potential of mobile technology to improve patient–provider communication, education, treatment, and adherence. In a study by Anderson et al. (2018), a mobile health intervention for youth with SCD improved their disease-related knowledge and functioning, as well as parent functioning. Testing such interventions could be a feasible and promising direction for research across contexts.

Conclusion

This study identified the types and magnitudes of burden in a population of caregivers of children with SCD in Kenya. Results suggest that caregivers experience significant difficulties related to their daily activities and their physical, social, cognitive, and emotional QOL. Given these findings, it is concerning, but not surprising, that many reported significant depressive symptoms. Financial hardship was reported and found to be a significant predictor for psychosocial burden across domains. Results can help guide the formation of ideas and hypotheses for intervention research in pediatric psychology for this population.

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