Psychosocial Burden of Childhood Sickle Cell Disease on Family Members and

Caregivers in Homa Bay, Kenya.

by

Bethany G. Kuerten

Duke Global Health Institute
Duke University

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Approved:

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Eve S. Puffer, Supervisor

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Amy H. Herring

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Melanie Bonner

Thesis submitted in partial fulfillment of
the requirements for the degree of
Master of Science in the Duke Global Health Institute
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ABSTRACT

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Abstract

Objective To characterize the types and magnitude of family burden present in caregivers and families who have a child with Sickle Cell Disease (SCD) in Kenya; to identify demographic and disease-specific predictors of caregiver burden. **Methods** Primary caregivers (N = 103) of children diagnosed with SCD completed surveys assessing multiple domains of caregiver quality of life, parental adjustment related to child illness, mental health, and financial burden. Descriptive statistics characterize caregiver burden and linear regressions models assess associations. **Results** On indicators of quality of life (QOL), caregivers report problems across most domains assessed included their daily activities and their 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 largest concern, though they also experience long-term uncertainty and sorrow and anger; they reported relatively high levels of emotional resources. Financial burden was high; caregivers reported moderate to major loss in revenue and financial benefits due to the time spent caring for the child. Linear regression analyses revealed that financial burden was a significant predictor of all negative outcomes. **Conclusions** Results provide preliminary evidence that Kenyan caregivers of children with SCD experience difficulties across multiple domains of functioning and that financial struggles are associated with most psychosocial outcomes. Results can begin to guide intervention needs and opportunities.

Key Words*: sickle cell disease, caregiver burden, chronic illness, Kenya*

Dedication

I dedicate my thesis work to Mom, Dad, and Nini. I wouldn’t be where I am or who I am without you.

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Acknowledgements

First, I would like to express my sincere gratitude to my advisor, Eve, for her support of my study, for her patience, motivation, immense knowledge, and kindness. Thank you to the Kenyan team—Sarah, Koech, and Edna—for accepting and teaching me while I explored and learned from the beautiful country and people around me in Kenya. Thank you to my participants as I wouldn't have been able to contribute this piece of work without them. Lastly, thank you to my classmates. I made lifelong friendships here and wouldn't have survived this program without the love, support, and laughs we shared along the way.

1. Introduction

1.1 Background

Sickle Cell Disease (SCD) is one of the most common genetic disorders of the blood (Edwards et al., 2005). This disease produces abnormal hemoglobin molecules in the red blood cells (RBCs) resulting in rigid, sticky, sickle-shaped cells (Edwards et al., 2005). SCD is a chronic condition that can cause painful episodes and symptoms resulting in progressive damage to multiple organ systems, and it often worsens with age (Anie, 2005; Rees, Williams, & Gladwin, 2010). Pain crises 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 December 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 impact of the disease, relatively little attention has been given to assessing the burden on children and families in Africa (Grosse et al., 2011).

It is estimated that the global number of neonates born with HbS include 5,467,000 carriers and 312,000 homozygous individuals, most of which occur in low- and middle- income countries (LMICs) (Piel, Patil, et al., 2013). Approximately 70% of SCD births occur in Africa (“WHO Global epidemiology,” 2008). Particularly for Homa Bay, Kenya, the HbS allele frequency ranges from 12-15% prevalence in the population (Malaria Atlas Project, 2010). In many LMIC contexts, health care resources and methods for early detection of SCD are limited, leading to undiagnosed and untreated

SCD that often results in early childhood mortality (Piel, Hay, Gupta, Weatherall, & Williams, 2013; Piel, Patil, et al., 2013). It is estimated that 50-90% of infants born with SCD in Africa die before age 5 (McGann, Hernandez, & Ware, 2017). However, despite these bleak statistics, there has been some encouraging shifts in SCD outcomes. As LMICs progress economically, there have also been some slow advances in healthcare, including care for SCD. Two main advances include both neonatal screening and the use of Hydroxyurea (HU) becoming more widely used across Africa, including in Kenya (Odunvbun, Okolo, & Rahimy, 2008; Tshilolo et al., 2019; Tusuubira et al., 2018). As a consequence of these changes, some patients with SCD have increased life expectancy and are surviving until reproductive age. With these achievements, there is now a clear need to consider the psychosocial dimensions of the illness to promote quality of life for children living with SCD and their families in LMICs (Ohaeri & Shokunbi, 2002). The field of pediatric psychology is well-positioned to make global contributions and understand the issues surrounding chronic conditions, like SCD, across many cultures and contexts.

SCD can place significant psychosocial burden on the individual child and larger family system (Barakat et al., 2007; Logan, Radcliffe, & Smith-Whitley, 2002). Children with SCD are affected in multiple ways; they may be held back from recreational activities, have school absenteeism due to frequent hospitalizations, have delayed puberty, experience decreased QOL, and have other limitations in social functioning (Hildenbrand, Barakat, Alderfer, & Marsac, 2015). In addition to these child-level stressors, childhood chronic illness often impacts entire family systems (Kazak, 1989). Caring for a child with SCD leads to physical and psychological demands, including facing overwhelming news of diagnosis, the associated medical risks and costs, the time required to attend medical appointments, and in many cases, the potential for shortened life expectancy (Cousino & Hazen, 2013). Parents of youth have been shown to have heightened psychological symptoms such as increased rate of depression and anxiety as well as socioeconomic hardship (Anie, 2005; Palermo, 2008; Sogutlu, Levenson, McClish, Rosef, & Smith, 2011) . A study in the Netherlands found that female family members caring for children with SCD exhibited worse health related QOL on all subscales, including depressive moods, disturbances in daily activities, vitality, sleeping, happiness, and cognitive functioning (van den Tweel et al., 2008). Madani and colleagues (2018) found that the majority of families had unmet financial needs and that many were experiencing high rates of exhaustion. In addition to these impacts, greater symptoms of distress in the parent has been linked to diminished QOL for the child (Panepinto, Hoffmann, & Pajewski, 2010).

While there is a growing body of literature examining the burden of caring for a child with SCD in developed countries, there are only a few studies in developing countries, particularly in African countries where the disease is prevalent. Similar to findings in developed countries, however, the few studies published among parents in African countries demonstrate significant psychosocial burden and financial stress. One study in Nigeria examined the psychosocial family burden of SCD, positing that the impact on family is worse in low-income countries due to inadequate social welfare and health care services. They found that family members of children with SCD experience significant financial stress, depressive symptoms, and neglect from other family members (Adegoke & Kuteyi, 2012). In Cameroon, researchers also found that the majority of participants experienced moderate to severe difficulty coping with having a child with SCD (Wonkam et al., 2014a). In Kenya, one study documented there was high levels of stigmatization surrounding the diseases resulting in loss of livelihood for mothers due to the limited care options (Marsh, Kamuya, & Molyneux, 2011). A key difference from the US findings is that parents in African countries report lack of national provisions and health care services, as well as greater stigmatization impacts (Adegoke & Kuteyi, 2012; Ohaeri & Shokunbi, 2002).

1.2 Study Aims

Despite the high prevalence of SCD in Africa, to date there are no studies to our knowledge examining the psychosocial sequelae of caring for a child with SCD in Kenya. The study aims to examine caregivers' perspectives on how having a child with SCD impacts functioning across multiple domains. We first aim to characterize the types and magnitude of burden present on individual caregivers and families. Secondly, we aim to identify demographic and disease-specific predictors of caregiver burden. Results can begin to provide the information needed for the development of interventions to reduce the impact of SCD on families (Piel, Patil, et al., 2013).

2. Methods

2.1 Setting

The study took place in Homa Bay, Kenya at the Homa Bay County Hospital. Homa Bay is located in Western Kenya on the border of Lake Victoria and has an estimated population of 965,000. In 2013, the poverty rate was estimated at 44% in the county. In addition, there is a high prevalence of SCD and incidence of malaria around the Lake Victoria basin. The combination of these diseases produces high risk for children, as children with SCD are more likely to die from malaria parasites. Malaria also often increases sickle cell pain, although the mechanisms of action are unclear (Fleming, 1989). Homa Bay is holoendemic for P. falciparum malaria, meaning that in the entire population is at risk of, or already infected with, P. falciparum malaria. In addition, the allele frequency for Sickle Cell Disease (HbS) is drastically high with 12-15% of the population with the HbS allele (Malaria Atlas Project, 2010). The Sickle Cell Clinic in Homa Bay is operated by a collaboration between Academic Model Providing Access to Health Care (AMPATH) HematoOncology and Moi Teaching and Referral Hospital (MTRH) located in Eldoret, Kenya. The clinic was established in 2013, and currently provides care to 1,400 patients, of whom 67% are under 10 years old. Clinic-based care includes treatment for pain crises and provision and prescription of HU. Other services, including neonatal screening and prophylactic penicillin, are available at Moi Teaching and Referral Hospital in Eldoret, Kenya, about 200 km from Homa Bay County Hospital.

2.2 Participants and procedures

Participants were recruited as part of a larger clinical trial, EPiTOMISE (Enhancing Preventative Therapy of Malaria in children with Sickle cell anemia in East Africa). EPiTOMISE aims to compare the efficacy of malaria medication to prevent P. falciparum malaria in children with Sickle Cell anemia. Any caregiver of a child with SCD eligible for EPiTOMISE was eligible to participate in this substudy. Inclusion criteria for these children included age greater than 12 months and less than 10 years at enrollment; current attendance at or willingness to attend the clinic; residence in specific nearby countries; confirmed hemoglobin genotype of HbSS by electrophoresis; no plans to relocate residence from these areas in the next 2 years; ability and willingness to take oral medication and adhere (or facilitate child adherence) to medication regimen; and the ability and willingness of parent or legally authorized representative (LAR) to consent. Exclusion criteria consisted of taking routine antimalarial prophylaxis for another indication; known chronic medical condition other than SCA requiring frequent medical attention; and/or receipt of a transfusion of red blood cells in the 120 days prior to screening. Participants were recruited between January and October 2018.

As participants of EPiTOMISE reported to the clinic for their clinical trial visits, the caregivers were approached by local research assistants who were fluent in English and two languages spoken in this region—Swahili and Dholuo. They were asked to participant in this sub-study. The study was described as a study about how caring for a child with SCD affects caregivers and families. After obtaining signed informed consent from the caregiver, interviews were conducted in the language most comfortable for the participant. All study procedures were approved by institutional review boards of Duke University and Moi Teaching and Referral Hospital.

2.3 Sample size calculation

Sample size was calculated using the power function in R (Champely, 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. The final sample included 103 individuals, increasing our power to 90% to detect this same effect size.

2.4 Measures

2.4.1 Item adaptation and development

Before administering survey instruments, items were translated from English to Swahili and Dholuo and then back translated to check for clarity. We began by conducting formative data collection to adapt the measures, as they were originally constructed for use in the United States. To do this, we conducted structured interviews to assess each item and free-listing exercises to generate potential new items. Both are detailed below.

2.4.2 Structured item assessment interviews

Eleven structured interviews were held with pairs of participants (N = 22). All survey items were assessed for comprehensibility, acceptability, and relevance to the community using van Ommeren's technique for transcultural translations (1999). These assess whether the item is understandable to the local population, whether people would feel comfortable answering the question, and whether the item is relevant to the local culture. If items did not meet each of these criteria, the participants were asked to provide suggestions. Items were revised and presented to subsequent groups until the item was understandable, appropriate and relevant. This process was repeated until all items met all criteria. For example, when the item "I feel sick to my stomach" was presented to the participants, they said the wording was not comprehensible and suggested it be changed to "I feel pain in my stomach." This revised item was then presented to a new group of participants and deemed comprehensible. Results led to nine items being revised.

2.4.3 Item development free-listing

In addition to the survey items being checked for cultural appropriateness, 21 participants were asked to do a free listing exercise to determine whether there were other items that should be added for this context. Free-listing included asking the participants to list anything that was not covered in the items that caused them stress and/or hardship. The purpose was to add any items relevant to this context that were not in the original measures developed in other settings. They were also requested to list any "good" things that come from having a child with SCD in their current situation to assess any positive or growth-oriented aspects of the caregiving experience, as some of these have emerged in the broader literature (Hullmann, Fedele, Molzon, Mayes, & Mullins, 2014). These responses were recorded and those that reflected the same constructs were grouped together. If 3 people mentioned similar topics, the item was added to the questionnaire as a "locally-developed item." Based on these results, nine questions were constructed that were specific to the local community that were not covered by items already included in the survey. These included four local items related to indicators of burden (alpha = 0.50), as well as five items pertaining to positive caregiving experiences (alpha = 0.48). Each of these items were structured to match the formatting of the questions already included in the survey.

2.4.4 Standardized Surveys

Four questionnaires were administered: The Pediatric Quality of Life (PedsQL) Family Impact Module (Varni et al., 2004), the Parent Experience of Child's Illness (PECI) Scale (Bonner et al., 2006), Financial Burden Questions (Adegoke and Kuteyi, 2012), and the Patient Health Questionnaire-2 (PHQ-2) (Kroenke et al., 2003).

The Pediatric Quality of Life (PedsQL) Family Impact Module (FIM)

The PedsQL FIM is designed to measure the impact of pediatric chronic health conditions on parents and the family (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004). This is a 36-item questionnaire that encompasses 7 subscales measuring parent self-reported functioning across multiple domains: Physical Functioning (6 items; α= 0.86), Emotional Functioning (5 items; α=0.71), Social Functioning (4 items; α=0.71), Cognitive Functioning (5 items; α=0.81), Communication (3 items; α=0.39), Worry (5 items; α=0.69), Daily Activities (3 items; α= 0.85), and Family Relationships (5 items; α=0.75). A 5-point scale is used for each item: 0, "Never a problem"; 1, "Almost Never"; 2, "Sometimes"; 3, "Often"; 4, "Almost Always." Items are reverse-scored and linearly transformed to a 0-100 scale (0=100, 1=75, 2=50, 3=25, 4=0) such that higher scores indicate better functioning. Three total scores were calculated from these various domains. The PedsQL FIM Total Scale Score is the sum of all 36 items divided by the total number of items answered (α= 0.84). The Parent Health Related Quality of Life (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=0.88). The Family Functioning Score (8 items) is computed as the sum of items divided by the number of items answered in the Daily Activities and Family Relationships Scale (α=0.21). Given the low internal consistency for Family Functioning, we did not use this scale for subsequent analysis.

The Parent Experience of Child's Illness (PECI) Scale

The PECI is a 25-item self-report measure of a parent's adjustment to their child's chronic illness. The measure includes a total of 25 items and four subscales, with some items loading onto multiple subscales. These include: guilt and worry (11 items; α=0.69), emotional resources (5 items; α=0.41), unresolved sorrow and anger (8 items; α=0.52), and long-term uncertainty (5 items; α=0.58). Each item is rated on a five-point Likert-type scale: 0, "Never"; 1, "Rarely"; 2, "Sometimes"; 3, "Often"; and 4, "Always." Scores for each subscale are calculated by adding the item responses and dividing by the number of items on that subscale. We also calculated a PECI composite score encompassing guilt and worry, unresolved sorrow and anger, and long-term uncertainty (α=0.69).

Financial Burden Assessment items

In order to address the impact on family finances, a set of questions were adapted from a study in Nigeria assessing the psychosocial burden of SCD, including financial hardship (Adegoke & Kuteyi, 2012). Four items were administered to ask about the extent to which their child's SCD has impacted their household financially: has the family lost revenue, has the time spent caring for the child impacted financial benefits, has the money spent impacted the family, and has the family taken out loans or sold valuable property to meet the expenditure on the child's illness. These were rated on a scale from 0, "No Loss"; 1, "Minor Loss"; 2, "Moderate Loss"; and 3, "Major Loss."

These were summed and divided by the total number of questions for a total financial burden assessment score. Families were also asked who is primarily responsible for settling the hospital bills. This was a multiple-choice question with answer choices "Mom/Dad," "Grandparents," "Other family members," and/or "Friends."

The Patient Health Questionnaire- 2 (PHQ-2)

The PHQ-2 includes two questions about the frequency of depressed mood and anhedonia, or the inability to feel pleasure, over the previous two weeks. The score ranges from 0-3 as follows: 0, "Not at all"; 1, "Several days"; 2, "More than half the days"; 3, "Nearly every day." The total PHQ-2 score can range from 0-6, with scores of 3 points or above indicating a major depressive episode according to US norms. The PHQ-2 has been used and validated in a study of HIV/AIDS and depression in Western Kenya (Monahan et al., 2009).

Demographics and Disease Severity

Demographic data were collected as part of the larger study through caregiver self-report. Disease severity was evaluated by examining the number of pain crises and number of hospitalizations experienced in 12 months as reported by caregivers.

2.5 Data Analysis

Data were analyzed using Excel and R version 3.5.1 (2018). Descriptive statistics were calculated to characterize the levels of family burden. Multiple linear regression was used to determine whether the outcome variables—indicators of caregiver burden—were associated with potential predictors: child age, child gender, number of pain crises, and level of financial burden.

3. Results

3.1 Participant Characteristics

Participants included 103 primary caregivers of pediatric patients diagnosed with Sickle Cell Disease. The majority of caregivers were mothers (72.65%). Most caregivers had only completed primary school (42.72%) and half worked outside the home (50.49%).

The average age of children was 5.39 years with a range of 12 months to 10 years. All except one child over the age of 5 was enrolled in school. Among the 72 children enrolled in school, they missed an average of 4.10 days of school in a 30-day period. Nearly 43% of the children had been hospitalized 1 or more times in the past year due to complications associated with SCD. Of the child enrolled in the study, 60.19% of them were using Hydroxyurea as the primary medication to treat SCD. Lastly, 20 of the children have or had another sibling with SCD, and 9 of those died due to the disease.

Table 1: Demographic Characteristics

	M	*SD*
Age of child (n= 103)	5.39	2.61
Child Sex	*n*	%
Female	39	37.86
Child Education Levels		
Not in class	31	30.10
Nursery	40	38.83
Class 1	13	12.62
Class 2	10	9.71
Class 3	6	5.83
Class 4	1	0.97
Class 5	2	1.94
Number Pain Crises (past 12 months)		
0 times	17	16.50
1 time	13	12.62
2 times	12	11.65
3-5 times	43	41.75
6-10 times	9	8.74
More than 10 times	9	8.74
Caregiver Relationship to Child		
Mother	84	81.55
Father	17	16.50
Aunty	1	0.97
Grandparent	1	0.97
Caregiver Education Levels		
No Education	2	1.94
Primary	44	42.72
Some Secondary	15	14.46
Secondary Completed	23	22.33
College	20	19.42
Caregiver Employment Status		
Outside the home	52	50.49
Inside the home	51	49.51

Note. Pain crisis is defined as pain lasting 2 hours or more that did not have an obvious cause. Number of pain crises the child experienced in the past 12 months.

3.2 Aim 1: Types and Magnitude of Burden

The descriptive information on each of indicator of burden is described below.

See Table 2 for full results.

Table 2. Mean and standard deviation scores of standardized surveys

Scales	*M(SD)*
PedsQL 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	62.30 (22.92)
Worry	47.91 (23.97)
Daily Activities	25.00 (25.99)
Family Relationships	58.50 (25.17)
Parent HRQOL Score	39.09 (22.37)
Total Score	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 (+)	2.59 (0.72)
Composite Score	1.98 (0.57)
Finances (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 Pos. Exp. 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)
PHQ2 (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. The Total Impact score is computed by averaging all 36 items. The HRQOL summary score is computed by averaging the 20 items comprising the Physical, Emotional, Social, and Cognitive Functioning subscales. Higher values indicate better HRQOL and family functioning. Composite PECI Score combines Guilt and Worry, Unresolved Sorrow and Anger, and Long-term Uncertainty. Finances survey scored with higher scores indicating worse functioning.

3.2.1 Quality of Life

Participants reported difficulties across the majority of quality of life domains. This is reflected in the overall Parent HRQOL and the Total composite scores with the sample population responding, on average, that they “Sometimes” or Almost Always” experience problems across the subscales. Participants reported the lowest scores on the Daily Activities subscale, reflecting family activities taking more time and effort, difficulty finding time to finish household tasks, and feeling too tired to finish chores around the house. On average, participants’ responses suggested that many “Often” or “Almost Always” experienced these types of problems. Participants also reported lower QOL on the Physical, Emotional, Social, and Cognitive domains. On average, they reported “Sometimes” or “Often” for items on these subscales. While mean responses are likely more informative, we also examined single items to identify specific problems that were heavily endorsed by the participants. We highlight some of those here. On the Physical subscale, the vast majority reported “Often” for feeling tired during the day. On Emotional, the majority reported “Often” to “Almost Always” feeling anxious. On Social Functioning the majority endorsed difficulty finding time for social activities “Often” to “Almost Always.” Lastly, for Cognitive, the vast majority reported “Often” finding it hard to keep their attention on things.

The highest quality of life scores—or fewest problems—were on the Family Relationships and Communication scales. On Family Relationships, single items showed that participants reported *not* often experiencing problems related to the lack of communication between family members or conflicts between family members. On the Communication Scale, participants reported very low difficulty communicating with doctors and nurses or talking about their child's health with other people in the community.

Two local indicators related to quality of life focused on experiencing outside stigma or having people avoiding them due to the illness. On average, respondents indicated that they "Rarely" to "Sometimes" had these experiences.

3.2.2 Parent Adjustment

On the PECI, higher numbers indicate increased experienced burden, except on the Emotional Resources subscale. As seen in Table 1, participants reported the highest scores on the guilt and worry subscale, with mean scores reflecting that they "Sometimes" or "Often" experience these. In particular, many caregivers endorsed waking up during the night to check on their child, worrying that something bad will happen when they are out of the caregiver's care, as well as worrying that when the child is playing, they will get hurt. Long-term Uncertainty and Unresolved Sorrow and Anger scores are similar, indicating that caregivers frequently experience thoughts and distress within these domains as well. Within the Long-term Uncertainty subscale, single item analysis showed particularly high endorsement of worrying about whether their child will be able to live independently as an adult. Caregivers also indicated that they "Sometimes" experience thoughts and feelings related to ongoing fear of possible illness consequences and long-term sadness. Emotional Resources items showed a more positive result. Caregivers' scores reflected that many feel aware of the particular ways that they react to sadness and loss, and that they are aware that they can get support when they need it. Most caregivers reported that they were "almost always" ready to face challenges related to their child's well-being. The majority also reported that they hardly ever feel jealous of parents who have healthy children and that seeing healthy children makes does not make them feel sad.

3.2.3 Financial Burden

Families and caregivers were most impacted financially as it pertained to losing revenue due to instances of losing a job, losing working hours for business, or missing opportunities to go to the farm or market to sell. Related, they also reported losing general financial resources due to time spent caring for the child with Sickle Cell Disease. On both of these indicators, caregivers, on average, reported "Moderate Loss" to "Major Loss" related to these problems. While these were the two most frequently endorsed problems, many caregivers also reported problems related to having spent money due to the child's illness. On whether spending on expenses related to the actual illness has affected the family, mean responses suggested that families were "affected but not significantly" or "affected but can cope." When asked specifically whether they had to take out loans or sell property to cover medical expenses, they on average, reported that this occurred between small and moderate amounts; very few reported "large sums which they may not be able to repay." When asked who settles the hospital bills for the child, 89% indicated that the mother or father is responsible.

Two locally-developed indicators also related to access to resources. The sample population, on average, reported "Rarely" to "Sometimes" experiencing challenges with accessing medications and worrying about getting to a clinic or hospital for treatment during a pain crisis.

3.2.4 Local Positive Experience Indicators

Participants were also asked to respond to local positive experience indicators. The sample population highly endorsed having all five positive experiences assessed by the questions, including feeling happy that there is better access to medications and better equipped to care for other children and family members, happiness surrounding creating awareness about the disease, and confidence about their child's survival and plans for the future. The average participant responded "Often" and "Always" to all of these items.

3.2.5 Depression

The mean score on the PHQ-2 was 3.74. The majority, 69.53%, of caregivers scored a 3 or above, indicating likely symptoms of depression.

3.3 Aim 2: Predictors of Caregiver Burden

Linear regression models were generated with the each of indicators of burden as the outcome and child age, child gender, finances score, and number of pain predictors as independent variables. The results are described below. See Table 3 for full results.

Table 3: Linear Regression models: Predictors of caregiver burden

Variable	Adjusted R2	df	F-statistic	Estimate	Estimate P-value	
PedsQL FIM						
Parent HRQOL Score	0.32	4, 98	13.1			
Child Age				0.88	0.22	
Child Gender				0.47	0.90	
Finances Score				-20.17	8.28E-10	*
Number of Pain Crises				-2.03	0.10	
Total Score	0.35	4, 98	14.83			
Child Age				0.90	0.10	
Child Gender				-0.72	0.81	
Finances Score				-16.62	7.15E-11	*
Number of Pain Crises				-3.73	0.11	
Physical Score	0.21	4, 98	7.799			

Child Age				0.71	0.45	
Child Gender				0.65	0.90	
Finances Score				-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	
Finances Score				-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	
Finances Score				-23.57	8.56E-10	*
Number of Pain Crises				-1.72	0.24	
Cognitive Score	0.13	4, 98	4.714			
Child Age				0.58	0.55	
Child Gender				-2.01	0.70	
Finances Score				-17.29	5.54E-05	*
Number of Pain Crises				-1.23	0.47	
Communication Score	0.058	4, 98	2.564			
Child Age				0.74	0.39	
Child Gender				0.52	0.91	
Finances Score				-8.02	0.03	*
Number of Pain Crises				-3.07	0.04	*
Worry Score	0.07	4, 98	2.898			
Child Age				0.33	0.71	
Child Gender				-0.86	0.86	
Finances Score				-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	
Finances Score				-21.82	1.67E-08	*
Number of Pain Crises				-1.17	0.43	
Family Relationships Score	0.04	4, 98	2.092			
Child Age				0.59	0.54	
Child Gender				-8.41	0.10	
Finances Score				-10.19	0.01	*
Number of Pain Crises				0.96	0.56	
PECI						
Composite Score	0.14	4, 97	5.285			
Child Age				-0.03	0.19	
Child Gender				0.06	0.59	
Finances Score				0.37	3.16E-05	*
Number of Pain Crises				0.03	0.34	
Guilt and Worry	0.18	4, 98	7.876.658			
Child Age				-0.04	0.11	
Child Gender				-0.14	0.25	
Finances Score				0.43	1.47E-05	*
Number of Pain Crises				0.01	0.71	
Emotional Resources	0.02	4, 98	1.62			

Child Age				0.07	0.01	*
Child Gender				-0.08	0.59	
Finances Score				-0.19	0.10	
Number of Pain Crises				-0.01	0.90	
Unresolved Sorrow/Anger	0.06	4, 98	2.768			
Child Age				-0.02	0.43	
Child Gender				-0.02	0.90	
Finances Score				0.31	0.003	*
Number of Pain Crises				0.04	0.37	
Long-Term Uncertainty	0.06	4, 98	2.755			
Child Age				-0.02	0.44	
Child Gender				0.34	0.05	*
Finances Score				0.37	0.01	*
Number of Pain Crises				0.05	0.37	
Depression						
PHQ2 Score	0.21	4, 97	7.68			
Child Age				-0.03	0.63	
Child Gender				0.11	0.77	
Finances Score				1.55	3.05E-07	*
Number of Pain Crises				-0.05	0.64	

Note. df, degrees of freedom; Total Impact score is computed by averaging all 36 items. HRQOL is computed by averaging the 20 items comprising the Physical, Emotional, Social, and Cognitive Functioning subscales. Higher values indicate better HRQOL and family functioning. Composite PECI Score combines Guilt and Worry, Unresolved Sorrow and Anger, and Long-term Uncertainty. Finances survey scored with higher scores indicating worse functioning.
*p<0.05

3.3.1 Quality of Life

Composite scores related to multiple domains of quality of life, as assessed by Total PedsQL FIM and Parent HRQOL scores, were associated significantly with financial burden. As expected, higher financial burden is associated with lower overall QOL for both composite scores. Likewise, for all subdomains, higher financial burden was associated significantly with worse QOL. Higher number of pain crises also emerged as a significant predictor of Physical, Emotional, and Communication QOL scores, with more crises associated with lower QOL. Pain crises were not associated with scores on the Social, Cognitive, Worry, Daily Activities, and Family Relationships subscales at an alpha level of .05, though all except for Family Relationships was in the hypothesized direction. Child gender did not predict any PedsQL FIM subdomains. Child age was associated significantly only with Daily Activities, indicating that as a child get older, burden related to daily activities increases.

3.3.2 Parental Adjustment

Financial burden was a significant predictor of the composite PECI score. As expected, higher financial burden was associated with greater parental distress. For subscales, higher financial burden was associated significantly with Guilt and Worry, Unresolved Sorrow and Anger, and Long-term Uncertainty, but not with Emotional Resources. Child age emerged as a significant predictor for Emotional Resources, with higher age associated with higher Emotional Resources. Pain crises and child gender were not associated with any of the subdomains at an alpha level of .05.

3.3.3 Caregiver Mental Health

Financial burden was associated significantly with symptoms of depression as assessed by the PHQ2. Higher financial burden was associated with higher endorsement of having little interest in doing things and feeling down or hopeless. No other potential predictor was associated with these symptoms.

4. Discussion

The current study sought to provide an understanding of caregiving stress and burden associated with pediatric SCD in Kenya. This adds to the very few studies characterizing the types and magnitude of caregiver burden in LMICs, and Africa specifically. Results showed that families experience difficulties related to multiple domains of QOL and adjustment to their child's illness. Participants reported particularly high burden in regard to very practical struggles in daily activities, such as difficulty finding time to finish household tasks and feeling too tired to complete tasks. In regard to parental adjustment to SCD, parents endorsed generally high distress overall. Of the areas assessed, guilt and worry was found to be particularly elevated among these caregivers. Participants also faced financial burden related to losing revenue due to loss of job and working hours. Lastly, more than half of caregivers reported significant symptoms of depression.

Despite experiencing many types of distress, caregivers did indicate some positive experiences, as they reported not feeling stigmatized in the community and feeling at peace with their circumstances. In addition, these families reported strong communication and family relationships. They reported little conflict between family members. Caregivers also reported that they communicate well with doctors and nurses, as well with others in the community.

The literature suggests some similarities and differences between US-based populations and this Kenyan sample, though without matched samples, comparisons must be made with caution. For example, Panepinto and colleagues (2009) evaluated caregiving burden in a population of pediatric sickle cell patients. They reported that caregivers reported significant burden related to worry and communication. Worry was also a concern in this Kenyan population, but communication was actually one of the areas of least concern in this sample. For the Kenyan families, communication with family and providers was the area with the highest reported functioning outcomes. Regarding parental adjustment to child illness, few studies of SCD have used the same assessment measure as the current study—the PECI. However, compared with a study using the PECI among parents of children with brain tumors in the US, many similarities were found. Findings related to long-term uncertainty and chronic sorrow were generally consistent across the two samples. However, the parents in Kenya of children with SCD did report more guilt and worry and slightly fewer emotional resources. These types of differences across settings may be related to cultural and contextual factors, which should be a focus of future research guiding whether intervention approaches helpful for parents in the US may be applicable in other settings.

The second aim of the study was to examine potential predictors of these levels of burden among caregivers. Results showed that financial burden is associated significantly with poorer outcomes across all parental distress and quality of life domains. This suggests that financial stressors are likely having broad effects on the psychosocial well-being of these parents. It has been shown in the literature that adverse health shocks, especially those related to chronic conditions, have impacts on economic outcomes for households in low- and middle-income countries (LMICs) (Alam & Mahal, 2014). Poor health can increase the likelihood that families will become impoverished if there is significant out-of-pocket (OOP) payments for healthcare (Alam & Mahal, 2014). These risks can cause dramatically lower HRQOL and greater tendency towards psychological distress (Park & Look, 2018). Our results indicate similar findings. In another study addressing stigmatization around SCD in Kenya, it was identified that families, especially mothers, had relatively few options in coping with economic losses and faced hardship in maintaining income generating activities (Marsh et al., 2011). Our results also support these findings as caregivers were most impacted financially as it pertained to losing revenue due to instances of loss of job or loss of working hours. Future research could examine more closely these experiences of families to identify potential points of intervention in terms of reducing the burdens related to both time and finances.

More than half of the sample indicated signs of depression. Along with the significant difficulties in QOL in this population, results could involve a cycle where negative thoughts reinforce negative emotions, which in turn, produce maladaptive, or distress-reinforcing, behaviors. These are likely associated with, and compounded by, the financial pressure that these families face. A study conducted by Sil and colleagues (2016) discusses catastrophizing as a specific type of thought pattern that resulted in increased pain for the child and emotional distress for the parent. Further depression screening is needed to see if this is the case in the Kenyan population; if so, being aware of this negative thought pattern could help guide interventions. Integrating a referral system of care can also aid this community.

There were several strengths to the design and implementation of this exploratory study. First, and most importantly, this research fills a gap in SCD literature from LMICs, and Kenya specifically. Critical information was gathered from a new population that can aid in future interventions and spark interest in learning more about Kenya and other low-resource areas of the globe where SCD is prevalent. Secondly, multiple domains of parent distress were assessed to gain a multi-faceted understanding of these caregivers' experiences. While the measures were standardized in the US, they were carefully assessed for cultural and contextual language. All measures showed high internal consistency, which is a positive indicator related to expected validity of results.

With regard to limitations of this study, we administered the questionnaire to a group of parents within one illness population. Patients with SCD represents a distinct community in terms of treatment, mortality, and long-term physical and psychosocial sequelae. Generalizability is limited for other chronic conditions that may lead to different stressors. In addition, 102 participants are still a relatively small sample size that was strictly limited to one geographic area, so it is also unclear how generalizable the results for other parts of Kenya or other LMICs. More research is needed with larger populations and in other locations.

The results of this study help determine the future directions and feasibility of interventions in this community. First, it is critical to recognize the financial burden that these caregivers are facing when considering any psychosocial programming, as well as any systems-based or policy initiatives. Possible intervention approaches could include training medical professionals on how to address the psychosocial burden that accompanies chronic disease, even in short clinical encounters. For example, they could provide very brief psychoeducation for caregivers on how childhood SCD can affect the family and create an environment that normalizes and validates the stress caused by the illness. Success on this front has been shown in Uganda by the Uganda Sickle Cell Rescue Foundation. The Foundation has been working with the community to break down barriers to care and decrease stigmatization around the disease. The Foundation also indicates that it is essential for the inclusion of SCD in existing health education programs at both the community and health center levels (Tusuubira et al., 2018).

Another intervention approach could include sickle cell support groups for caregivers. Benefits of support groups include finding a shared social identity which fosters a sense of belonging, support, and empowerment for both patient and caregiver (Shilling et al., 2013). Support groups also allow for personal growth, the opportunity to learn from others, improvements in family functioning and psychological health (Shilling et al., 2013). In addition to this, there is the opportunity to capitalize on technology and use of mobile phones in the area. In Eldoret, a larger city located East of Homa Bay, their support groups consist of in-person meetings and community forums initiated through WhatsApp. This allows for faster transfer of knowledge and it is potentially feasible as it is already being done in a similar context. Technology-based interventions also hold great potential as they have the ability to reach large numbers, even in remote areas. Mobile technology also has the potential to improve communication between patients and medical providers to achieve more complete healthcare that encompasses education, treatment, and adherence (Anderson et al., 2018). In a 2018 study by Anderson and colleagues, a mobile health intervention was assessed to reveal its impact on adherence, disease knowledge, and quality of life in pediatric SCD patients. They found that participants experienced increased disease knowledge, significantly better SCD-related functioning, as well as higher parent functioning (Anderson et al., 2018). Additionally, McClellan and colleagues found that there was high participation rates and consumer satisfaction with home-based technology targeting behavioral coping skills in participants with SCD (McClellan et al., 2009). In conclusion, these examples of mobile programming could be implemented for the community in Homa Bay for the children and their caregivers.

5. Conclusion

This study identified the types and magnitudes of burden present in a population of caregivers who are caring for a child with sickle cell disease in Homa Bay, Kenya. This is one of few studies in LMICs on the psychosocial burden of this childhood illness. The results suggest that caregivers are impacted heavily in their daily activities and physical, social, cognitive, and emotional quality of life; many reported significant depression symptoms. Also, financial burden was particularly high and was a predictor for all negative types of burden. They did not, however, experience as much hardship in their family relationships or with communication among family members and medical professionals. Results provide preliminary evidence that Kenyan caregivers of children with SCD experience difficulties across multiple domains of functioning and that financial struggles are associated with most psychosocial outcomes. Results can begin to guide intervention needs and opportunities.

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