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## The Association Between Caregiver Coping and Youth Clinic Attendance and Health Related Quality of Life in Pediatric Sickle Cell Disease

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### Abstract

Caregivers of youth with sickle cell disease (SCD) influence youth disease management and psychosocial outcomes. Effective caregiver coping is important for improving disease management and outcomes since caregivers often report high disease-related parenting stress. This study characterizes caregiver coping and examines its relation to youth clinic non-attendance and health-related quality of life (HRQOL). Participants were 63 youth with SCD and their caregiver. Caregivers completed the Responses to Stress Questionnaire–SCD module to assess primary control engagement (PCE; attempts to change stressors or reactions to stress), secondary control engagement (SCE; strategies to adapt to stress), and disengagement (avoidance) coping. Youth with SCD completed the Pediatric Quality of Life Inventory–SCD module. Medical records were reviewed for hematology appointment non-attendance rates. Coping factors were significantly different ( $F(1.837, 113.924)=86.071, p<.001$ ); caregivers reported more PCE ( $M=2.75, SD=0.66$ ) and SCE ( $M=2.78, SD=0.66$ ) than disengagement ( $M=1.75, SD=0.54$ ) coping. Responses to short-answer questions corroborated this pattern. Greater caregiver PCE coping was associated with lower youth non-attendance ( $\beta=-0.28, p=.050$ ), and greater caregiver SCE coping was related to higher youth HRQOL ( $\beta=0.28, p=.045$ ). Caregiver coping is related to improved clinic attendance and HRQOL in pediatric SCD. Providers should assess caregiver coping styles and consider encouraging engagement coping.

### Keywords

sickle cell disease; coping; clinic attendance; health-related quality of life

### Introduction

Sickle cell disease (SCD) affects approximately 100,000 Americans and is the most common genetic disorder in the United States.<sup>1</sup> The treatment regimen for SCD is complex,

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necessitating that families complete preventative (e.g., hydroxyurea adherence) and reactive (e.g., pain management) self-management behaviors. To prevent impaired health and health-related quality of life (HRQOL), the National Heart, Lung, and Blood Institute (NHLBI) recommends that youth with SCD are seen every three to 12 months for Transcranial Doppler Ultrasonography (TCD), blood pressure, renal, and other screenings.<sup>2</sup> Patients are often seen even more frequently when initiating hydroxyurea (e.g., monthly<sup>3</sup>) or receiving chronic blood transfusions (e.g., once every 3-4 weeks<sup>4</sup>). Unfortunately, despite its importance for disease management, clinic attendance rates for pediatric SCD are between 46-77%,<sup>3,5</sup> and youth with SCD report significantly lower HRQOL across domains compared to healthy peers.<sup>6-8</sup>

Caregivers are often highly involved in youth SCD management, including monitoring and managing youth symptoms, administering medication, and facilitating clinic attendance in addition to other everyday parenting responsibilities.<sup>9</sup> The transactional stress and coping model<sup>10</sup> provides a framework to examine the relationship between caregiver functioning and youth outcomes in pediatric chronic illnesses. This model posits that adjustment to chronic illness is influenced by multiple demographic, disease, and family-related processes. Caregiver adaptational processes in this model include appraisal of disease-related stress and use of coping strategies.<sup>11,12</sup> Consistent with this model, caregiver functioning in pediatric SCD is associated with youth disease management predictors and outcomes.<sup>13,14</sup> For example, caregivers of youth with SCD report high levels of disease-related emotional<sup>15</sup> and parenting stress,<sup>9,16,17</sup> which is linked to lower HRQOL,<sup>14</sup> higher pain,<sup>16</sup> and more hospital utilizations<sup>18</sup> and urgent care use<sup>13</sup> in youth.

Effective caregiver coping has the potential to improve disease management and mitigate poor disease outcomes. Empirically validated frameworks of coping in pediatric chronic illness include three similar subtypes of coping that are grounded in Rothbaum, Weisz, and Snyder's control-based model of coping.<sup>19,20</sup> Primary control engagement (PCE) coping is characterized by attempts to change the source of stress directly (e.g., problem solving) or one's emotional reactions to the stressor (e.g., emotion expression, emotion modulation). This form of coping is considered most adaptive when addressing controllable stressors.<sup>20</sup> Secondary control engagement (SCE) coping reflects accommodative coping strategies, such as reappraisal, acceptance, distraction, or positive thinking; it is hypothesized to be most adaptive when managing uncontrollable stressors.<sup>20</sup> Finally, disengagement coping is characterized by attempts to avoid the source of stress, both behaviorally and cognitively.<sup>19</sup>

The impact of caregiver coping on youth outcomes in pediatric SCD is a developing area of research. Caregiver coping strategies, including problem solving and planning (PCE coping),<sup>21</sup> attempts to accommodate and adapt to stressors (SCE coping),<sup>22</sup> and avoidance (disengagement coping),<sup>23-25</sup> are associated with youth disease management measures and outcomes in numerous pediatric populations, including HIV,<sup>26</sup> spinal cord injury,<sup>23</sup> cystic fibrosis,<sup>24</sup> cancer,<sup>25</sup> and asthma.<sup>21,22</sup> Extant pediatric SCD literature shows that greater caregiver use of disengagement coping has been correlated with more youth internalizing symptoms<sup>27</sup> and youth pain-related utilization.<sup>28</sup> Additionally, higher caregiver cognitive and behavioral (PCE and SCE) coping strategies are associated with more youth involvement in social activities.<sup>29</sup> Further, literature suggests that frequency of coping

strategy use is modifiable via intervention, and such interventions lead to improvements in caregiver and youth psychosocial outcomes. For example, interventions targeting caregiver problem solving, a PCE coping strategy, led to reduction of caregiver distress, anxiety, depression, pain catastrophizing, and post-traumatic stress,<sup>30-32</sup> as well as improvements in youth depressive and anxious symptoms.<sup>30</sup> Additionally, a meta-analysis examining coping interventions in caregivers of hospitalized youth found that interventions significantly reduced parental anxiety and depression.<sup>33</sup> However, a better understanding of caregiver SCD-specific coping and its associations with both predictors and functional outcomes of disease management in pediatric SCD is critical.

There is limited research regarding the relationship between caregiver coping and both youth clinic attendance and HRQOL, which are important disease management measures/outcomes. Multiple variables are associated with clinic attendance (e.g., distance from clinics, insurance status<sup>34,35</sup>); however, effective caregiver coping may be a modifiable factor that has the potential to improve clinic attendance in pediatric SCD populations by addressing some of the documented protective (e.g., spirituality, social support) and risk (e.g., cognitive impairment, depression) factors for clinic attendance and non-attendance.<sup>36</sup> The current study aims to address these gaps by characterizing caregiver disease-specific coping and examining its relation to clinic non-attendance and disease-specific HRQOL. We hypothesized that caregivers would engage in higher rates of PCE and SCE coping compared to disengagement coping. Caregiver short-answer responses were examined to answer the following research question: “What coping strategies do caregivers use to manage the stressful aspects of having a child with SCD?” We also hypothesized that higher rates of caregiver PCE and SCE coping would be associated with fewer non-attended appointments and higher HRQOL, while higher rates of caregiver disengagement coping would relate to more non-attended appointments and lower HRQOL. Results will provide a more in-depth understanding of common caregiver coping strategies and help determine whether caregiver coping is an important intervention target for improving SCD management and associated outcomes.

## Materials and Methods

### Participants

Participants were youth ages 8-18 with a documented diagnosis of SCD and a parent or legal guardian who were enrolled in a larger cross-sectional study on sleep disturbances and neurocognitive functioning.<sup>37</sup> Consistent with the protocol for the larger study, dyads were not eligible to participate if: 1) youth had a diagnosis of a neurological or genetic condition unrelated to SCD that may affect neurocognitive functioning; 2) youth were currently hospitalized for SCD-related complications; 3) either the caregiver or the youth did not speak English; and 4) if the caregiver and youth had not spent at least one night sleeping in the same place in the previous month.

### Procedures

Participants were recruited from the Pediatric Hematology and Oncology clinic at a university-affiliated hospital from February through September of 2019. Study staff

administered all measures either at the participant's clinic appointment or at a scheduled study visit. Caregivers and eighteen-year-old youth provided informed consent, and youth under eighteen provided assent. All study procedures were approved by the institutional review board (IRB), and participants received compensation for their participation in the study.

## Measures

**Participant Characteristics**—Caregivers completed a measure about basic demographic information, including caregiver and youth age, gender, race, and ethnicity. Electronic medical records were also reviewed to collect youth SCD genotype, which was stratified into HbSS disease and HbS $\beta^0$  thalassemia versus HbSC disease and HbS $\beta^+$  thalassemia, as done in previous literature.<sup>28</sup>

**Caregiver SCD-Specific Coping**—Caregivers completed the self-report version of the Responses to Stress Questionnaire's (RSQ) SCD module.<sup>38,39</sup> The RSQ is a 57-item measure that assesses voluntary (coping) and involuntary (automatic) responses to stressors. The three coping factors (PCE, SCE, and disengagement coping) were used for analyses in this study. Questions are multiple-choice ( $N = 57$ ) and short-answer ( $N = 2$ ). For multiple-choice questions, respondents are asked to report how much they engage in each response when faced with the stress of having a child with SCD on a scale of 1 ("not at all") to 4 ("a lot"). Higher scores indicate greater use of specific coping factors. The measure also includes a list of twelve SCD-related stressors (e.g., "Having to go to the hospital or clinic so often"), and respondents are asked to indicate how stressful each has been for them over the past six months on a scale of one (not at all) to four (very). Short-answer questions ask participants to expand upon multiple-choice responses and include the following: "I do something to try to fix the stressful aspects of having a child with SCD (write one thing you did)" and "I try to think of different ways to change or fix the situation (write one plan you thought of)." These open-ended questions allow for more nuanced exploration of caregivers' coping strategies.

The standard method of scoring of RSQ quantitative data uses proportion scores, which are calculated by dividing each factor score by the total RSQ score. Proportion scores are used to control for response bias as well as individual differences in base rates of item endorsement; their use has been supported in the coping literature.<sup>40,41</sup> The RSQ has been used in SCD<sup>39,42</sup> and its factor structure has been supported by confirmatory factor analysis studies in a range of diverse samples.<sup>19</sup> The RSQ has also demonstrated acceptable psychometric properties in various disease populations.<sup>43,44</sup> Internal consistency for the overall measure was excellent ( $\alpha = 0.94$ ) in our sample; internal consistency for PCE, SCE, and disengagement coping subscales ranged from  $\alpha = 0.76$  (PCE) to  $\alpha = 0.80$  (disengagement).

**Youth Clinic Attendance**—Electronic medical records were reviewed to collect outpatient pediatric hematology clinic appointment attendance, including blood transfusion appointments, in the year prior to enrollment. A hematology/oncology provider and information technology specialist provided consultation regarding clinic attendance

categorization. Appointments were classified as attended, no-showed, canceled by the patient, canceled by a provider, or canceled due to patient hospitalization based on medical record descriptions. A percent non-attendance variable was calculated for main analyses by dividing the number of appointments no-showed and canceled by the patient by the number of appointments attended, no-showed, or canceled by the patient. The percent non-attendance variable captures the percent of missed appointments out of the total number of appointments patients had control over attending. This method of capturing clinic attendance outcomes as a percentage is similar to previous research.<sup>45-47</sup>

**Youth SCD-Specific Quality of Life**—Youth completed the SCD module of the Pediatric Quality of Life Inventory (PedsQL) to assess SCD-specific HRQOL.<sup>48</sup> Youth ages 8-12 completed the child-report version and youth ages 13-18 completed the teen-report version. This 43-item measure assesses SCD-related pain and hurting, pain impact, pain management and control, worrying, emotions, treatment, and communication. Participants report how much of a problem each item has been during the past month on a scale of 0 (never a problem) to 4 (almost always a problem). Total HRQOL scores and those for each subscale are calculated on a scale of 0 to 100, with higher scores indicating lower problems. The scale has been shown to be reliable, valid, and feasible in a pediatric SCD population.<sup>49</sup> Internal consistency for the overall measure was excellent ( $\alpha = 0.93$ ) in our sample; subscale internal consistency ranged from  $\alpha = 0.59$  (Communication 1) to  $\alpha = 0.89$  (Pain Impact).

## Data Analysis

Analyses were conducted using IBM SPSS, version 25.0. All but one participant, who was missing data from the PedsQL, had complete data. For analyses using HRQOL data, listwise deletion was used so the participant was removed from calculations. Descriptive statistics were used to characterize caregiver coping, youth HRQOL, and youth clinic attendance. A repeated measures ANOVA with a Greenhouse-Geisser correction using raw coping scores compared caregiver level of engagement in different types of coping as endorsed in multiple-choice responses. Post hoc comparisons used a Bonferroni correction. Partial eta squared ( $\eta^2$ ) was interpreted as a measure of effect size, with 0.01 indicating small, 0.06 medium, and 0.14 large.<sup>50</sup> For open-ended RSQ questions, three researchers independently reviewed and coded all caregiver responses. Researchers created a codebook with the definition of each code. Discrepancies between coders were resolved through discussion. After all data were coded, initial codes were collapsed into broader themes<sup>51</sup> and organized by coping subtype (PCE, SCE, or disengagement).<sup>52</sup> This organization of concepts using an existing theoretical framework is consistent with thematic survey analysis.<sup>52</sup>

A Pearson correlation test examined the relationship between potential covariates (child age, child gender, SCD genotype, family income, and caregiver education) and outcome variables. Potential covariates were determined based off previous literature in pediatric SCD, coping, and clinic attendance.<sup>28,36,53-57</sup> Two separate multiple linear regressions examined the association between the three caregiver coping proportion scores and percent non-attendance and SCD-specific HRQOL. All statistical assumptions for linear regressions were found to be tenable.  $R^2$  was interpreted as a measure of effect size, with 0.02 indicating small, 0.15 medium, and 0.35 large.<sup>58</sup>

## Results

### Descriptive and Preliminary Analyses

Sixty-three caregiver-youth dyads (66% of those approached and eligible) participated in the study. Mean youth age was 13.17, and the majority of youth were Black/African American ( $N = 61$ , 97%) and male ( $N = 38$ , 60%). Thirty-nine youth (62%) had HbSS disease, 12 (19%) had HbSC disease, 8 (13%) had HbS $\beta^+$  thalassemia, and 4 (6%) had HbS $\beta^0$  thalassemia. Eight youth (13%) were on chronic transfusions and 29 (46%) were prescribed hydroxyurea. See Table 1 for additional participant demographic and medical characteristics. Number of scheduled appointments per participant in the previous year ranged from 1 to 32 ( $M = 8.08$ ,  $SD = 5.85$ ; 25% had 5 scheduled appointments, 50% had 7 scheduled appointments, 75% had 9 scheduled appointments). On average, 32% of appointments were no-showed or canceled by the patient out of the total number of appointments they had the opportunity to attend (percent non-attendance dependent variable). Mean HRQOL score in this sample was 65.41; see Table 1.

**Caregiver Stress and Coping**—Average total caregiver SCD-related stress score was 2.46 ( $SD = 0.70$ ), indicating that SCD-related problems were “a little” to “somewhat” stressful and challenging to manage. The most stressful problems were “Not being able to help my child feel better” ( $M = 3.39$ ,  $SD = 0.98$ ) and “Not knowing if my child’s SCD will get better” ( $M = 3.06$ ,  $SD = 0.97$ ). Table 2 provides mean stress scores for each item. On average, caregivers engaged in PCE and SCE coping strategies “a little” to “some” of the time and used disengagement coping strategies “not at all” to “a little” of the time. See Table 2 for caregiver raw and proportion coping scores, as well as percentage of caregivers who use coping strategies at each level. There was a significant difference within the three coping factors ( $F(1.84, 113.92) = 86.07$ ,  $p < .001$ ,  $\eta^2 = .58$ ). Caregivers engaged in significantly more PCE ( $M = 2.75$ ,  $SD = 0.66$ ) and SCE ( $M = 2.78$ ,  $SD = 0.66$ ) coping strategies compared to disengagement coping strategies ( $M = 1.75$ ,  $SD = 0.54$ ) (both  $p < .001$ ); use of PCE and SCE coping strategies did not differ significantly ( $p = .72$ ). There were 73 total responses to both open-ended questions; these responses described a similar pattern of engagement in PCE (36 responses), SCE (38 responses), and disengagement (4 responses) coping strategies. Themes included caregiver engagement in PCE coping strategies such as prevention efforts, efforts to manage their child’s pain and/or treatment, problem solving, and SCD research, as well as caregiver engagement in SCE coping strategies such as distraction, relaxation, and/or engaging in pleasurable activities, religious/spiritual coping, and cognitive coping; see Table 4.

### Clinic Non-Attendance

There were no significant relationships between percent clinic non-attendance and potential covariates. Greater caregiver use of PCE coping strategies was associated with lower rates of non-attendance, and results approached significance ( $\beta = -0.28$ ,  $p = .050$ ). Caregiver SCE ( $\beta = 0.13$ ,  $p = .36$ ) and disengagement ( $\beta = -0.08$ ,  $p = .60$ ) coping strategies were not significantly related to percent non-attendance; see Table 3.



## HRQOL

There were no significant relationships between youth HRQOL and potential covariates. Greater caregiver use of SCE coping strategies was associated with higher youth HRQOL ( $\beta = 0.28, p = .045$ ). Caregiver PCE ( $\beta = 0.16, p = .26$ ) and disengagement ( $\beta = 0.04, p = .77$ ) coping strategies were not significantly linked to youth HRQOL; see Table 3.

## Discussion

Caregivers have an integral role in the lives of youth with SCD, but they frequently face a range of modifiable and non-modifiable stressors that can impede their ability to consistently help youth engage in self-management behaviors and promote optimal youth functioning. For example, caregivers in the current study reported “a little” to “somewhat” SCD-related stress, particularly surrounding not being able to help their child feel better and not knowing if SCD will get better. The current study aimed to characterize disease-specific coping with stressors in caregivers of youth with SCD and examine its relation to two key disease management measures/outcomes, youth clinic attendance and HRQOL.

Quantitative data from the current study suggest that caregivers engaged in significantly more PCE and SCE coping compared to disengagement coping. Caregiver responses to open-ended questions demonstrate a similar pattern of engagement in PCE and SCE strategies and highlight specific strategies used (e.g., prevention efforts, pain management, religious/spiritual coping). These qualitative responses provide a more in-depth understanding of caregiver engagement coping efforts in pediatric SCD. Engagement coping strategies are associated with better adjustment, while disengagement coping strategies have been related to poorer adjustment.<sup>19</sup> Thus, findings from the current study indicate caregiver resilience and use of adaptive coping strategies. These results are somewhat consistent with findings from a recent study of coping among caregivers of youth with SCD in Brazil,<sup>42</sup> although the study indicated lower proportion scores for caregiver PCE coping compared to the current study. Findings from the current study are also comparable to findings from other chronic illness populations; proportion scores for caregiver PCE, and disengagement coping in the current study are similar to RSQ coping proportion scores among caregivers of youth with asthma,<sup>22</sup> type 1 diabetes,<sup>59</sup> and cancer.<sup>55</sup> Extant literature indicates that coping-focused interventions can modify frequency of coping strategy use.<sup>30-33</sup> Interventions that focus on increasing PCE and SCE coping strategies lead to improved outcomes,<sup>30-33</sup> and such interventions are also shown to reduce use of disengagement coping strategies.<sup>60</sup> Thus, interventions in pediatric SCD should aim to help caregivers enhance current and build additional engagement coping strategies.

To our knowledge, this was the first study to examine the relationship between caregiver coping and youth clinic attendance in pediatric SCD. Contrary to hypotheses, caregiver PCE and disengagement coping were not related to youth HRQOL, but greater caregiver SCE coping was significantly associated with higher youth HRQOL as hypothesized. This is consistent with extant coping literature suggesting that SCE coping strategies such as acceptance and accommodation are associated with better adjustment.<sup>19</sup> SCE coping strategies are considered most effective when addressing uncontrollable stressors<sup>20</sup>; thus, perhaps SCE coping is helpful in improving HRQOL when facing uncontrollable or

less controllable aspects of SCD (e.g., pain). Findings provide preliminary evidence that caregiver SCE coping strategies such as distraction, acceptance, reappraisal, and positive thinking may be important intervention targets to promote youth HRQOL. It may be beneficial for providers to assess caregiver coping during clinic visits and encourage SCE coping responses. Evaluation of caregiver coping may also be an important target of psychological and social work consultation during hematology clinic appointments. Psychologists and social workers may consider providing psychoeducation about caregiver SCE coping strategies during routine consultations and referring families to outpatient therapy if warranted.

Also contrary to hypotheses, caregiver SCE and disengagement coping were not associated with youth clinic non-attendance. However, greater caregiver use of PCE coping was associated with fewer non-attended appointments, with results trending toward significance. PCE coping strategies such as problem solving and emotion modulation are hypothesized to be most beneficial when addressing controllable stressors,<sup>20</sup> and it is possible that these strategies may help reduce clinic non-attendance in pediatric SCD (e.g., by mitigating more controllable barriers). However, findings should be interpreted with caution due to non-significance. Existing interventions aimed at improving clinic attendance are limited,<sup>5</sup> but literature suggests feasibility and/or acceptability of in-person and web-based problem-solving interventions for both youth and caregivers to improve pediatric SCD clinic attendance and other self-management tasks.<sup>5,61</sup> More research is needed to continue to assess the impact of caregiver problem-solving and other PCE coping strategies on youth SCD clinic attendance.

The current study should be considered in the context of several limitations. Effect sizes in the current study were small-to-medium (e.g., explaining approximately 7% of the variance in youth clinic attendance and 12% of the variance in youth health-related quality of life), which suggests that other sociodemographic and clinical characteristics may be important contributors to youth disease outcomes. The transactional stress and coping model<sup>10</sup> postulates that multiple individual, caregiver, and family factors contribute to adjustment to chronic illness. However, the current study focuses solely on caregiver coping, which is one of many pathways in the model. It is possible that limited significant findings in the current study were partially due to the failure to account for additional individual (e.g., youth's own development and self-management) and clinical factors important to youth disease management predictors and outcomes. One important clinical factor that warrants more consideration is disease severity. Although SCD genotype (a proxy of disease severity<sup>28</sup>) was examined as a potential covariate and found unrelated to outcome variables, other measures of disease severity (e.g., hospitalizations, chronic transfusion status, experience of stroke, medication use) may contribute to youth outcomes.

The use of a comprehensive measure of caregiver disease-specific coping with both quantitative and open-ended responses is a strength, especially given the minimal research on caregiver coping with SCD-specific stressors. Still, the use of a self-report coping measure may have led to positive response bias. Although self-report coping measures are common in the literature,<sup>27,28</sup> the subjective nature of such questionnaires likely subjects them to biases, including selective recall bias and social desirability.<sup>62</sup> It is well-documented



in the literature that mistrust of behavioral and medical research in Black or African American communities may influence willingness to participate in research studies.<sup>63-65</sup> Thus, it is possible that potential mistrust of the medical and scientific community also contributed to participant response bias. Despite this potential bias, a strength of the current study is that only 6% of those approached and eligible for the study declined to participate (another 26% of those approached and eligible were unavailable but willing to be re-approached and then lost to follow-up; 1% [1 participant] withdrew from the study). Another limitation is the cross-sectional nature of the current study, which makes it difficult to determine the directionality of results or whether baseline caregiver coping predicts youth clinic attendance and health-related quality of life at follow-up. Additionally, participants were recruited during clinic visits, so the sample only included youth who attended at least one clinic appointment during the recruitment period and excluded youth that did not attend any visits. Number of scheduled visits varied widely in our sample, likely due to differences in SCD severity and treatment regimens (e.g., hydroxyurea, chronic blood transfusions). However, our categorization scheme did not account for differences between participants in total number of scheduled clinic appointments, so the impact of appointment regimen could not be addressed. Although electronic medical record review is an efficient and accurate method for capturing clinic attendance, there are some drawbacks to this approach. We were unable to distinguish between appointments that were canceled by the patient and rescheduled from those that were canceled with no rescheduled appointment.

Future studies should examine the relationship between caregiver coping and youth clinic attendance and HRQOL in larger SCD samples, which will increase power to detect potential additional effects of caregiver coping on youth outcome variables. In addition, prospective analyses should assess whether baseline caregiver coping predicts youth outcomes at follow-up. This will help researchers determine directionality of associations and identify the most salient intervention targets. Future studies should also examine whether there are any factors that are more directly impacted by caregiver coping that may, in turn, impact youth clinic attendance or health-related quality of life. These factors include additional sociodemographic variables, youth clinical characteristics (e.g., pain, disease complications, medications, treatment adherence), caregiver clinical characteristics (e.g., caregiver's own health and/or SCD diagnosis), caregiver and youth cognitive processes, family functioning, and sociocultural variables such as racism. SCD primarily affects Black individuals, and it is well-documented that racism and stigma have negative effects on youth SCD outcomes.<sup>66,67</sup> Caregiver experiences of racism may also impact caregiver coping efforts. Studies should also address other variables potentially associated with clinic attendance, such as families' distance from clinics, patient-provider relationships, insurance status, family responsibility sharing, and technology use.<sup>34,35,68</sup> Finally, in addition to evaluating statistical significance, future studies should assess what patients and caregivers would determine as the minimum reduction in scores necessary to produce results that are meaningful to them (i.e., the minimal clinically important difference [MCID]) in outcome measures such as the PedsQL-SCD.<sup>69</sup> Even if larger samples were to produce clinically significant findings, it is imperative to determine if associations are clinically meaningful. Findings should inform future clinical recommendations.

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TABLE 1

Participant demographics, medical information, and HRQOL

Variable	n (%) or $M \pm SD$	Range
Youth Age	13.17 $\pm$ 3.09	8.00-18.60
Youth Gender		
Female	25 (40)	
Male	38 (60)	
Youth Race and Ethnicity		
Black/African American	61 (97)	
Asian	1 (2)	
Hispanic or Latino/a	1 (2)	
Youth SCD Genotype		
HbSS or HbS $\beta^0$	43 (68)	
HbSC and HbS $\beta^+$	20 (32)	
Youth Hemoglobin	9.77 $\pm$ 1.92	2.68-13.50
Youth SCD-Related Treatment		
Chronic Transfusions	8 (13)	
Hydroxyurea	29 (46)	
Youth Stroke History		
Overt Stroke	3 (5)	
Silent Cerebral Infarction	7 (11)	
Youth Hospital Utilization		
Hospitalizations	0.70 $\pm$ 1.12	0-5
Emergency Department Visits	0.84 $\pm$ 2.29	0-17
Caregiver Age	39.30 $\pm$ 8.68	24-63
Caregiver Gender		
Female	57 (90)	
Male	6 (10)	
Caregiver Race/Ethnicity		
Black/African American	60 (95)	
Asian	1 (2)	
Hispanic or Latino/a	1 (2)	
Other	1 (2)	
Caregiver Relationship to Youth		
Mother	53 (84)	
Father	5 (8)	
Other <sup>a</sup>	5 (8)	
Caregiver Education: Highest Grade Completed	13.78 $\pm$ 2.42	11-20
Youth HRQOL		
Total Score	65.41 $\pm$ 17.28	18.60-99.42
Worry 2	79.37 $\pm$ 28.41	0-100
Communication 1	72.22 $\pm$ 25.53	0-100

Variable	n (%) or $M \pm SD$	Range
Worry 1	70.32 $\pm$ 26.24	0-100
Treatment	69.33 $\pm$ 20.97	21.43-100
Pain and Hurt	68.39 $\pm$ 20.43	19.44-100
Emotions	67.26 $\pm$ 31.94	0-100
Communication 2	61.64 $\pm$ 32.36	0-100
Pain Impact	55.85 $\pm$ 24.17	7.50-100
Pain Management	55.44 $\pm$ 31.58	0-100

Note. HRQOL = Health-Related Quality of Life.

<sup>a</sup>“Other” caregivers included 1 grandmother, 1 stepmother, and 3 legal guardians.

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TABLE 2

## Caregiver Stress and Coping Scores

Caregiver Stress and Coping	n (%) or $M \pm SD$
Total Stress Score <sup>a</sup>	2.46 ± 0.70
Not being able to help child feel better	3.39 ± 0.98
Not knowing if SCD will get better	3.06 ± 0.97
“Other”	3.00 ± 1.41
Effects of treatment	2.92 ± 1.15
Paying bills and family expenses	2.84 ± 1.17
Concerns about job or partner’s job	2.34 ± 1.27
Talking to others about SCD	2.21 ± 1.13
Needing more help/support from family & friends	2.16 ± 1.25
Talking to child about SCD	2.13 ± 1.08
Understanding information about SCD	2.05 ± 1.21
Arguing with child about treatment	2.02 ± 1.21
Having less time/energy for others	1.92 ± 1.08
Proportion Coping Scores	
PCE Coping	0.21 ± 0.05
SCE Coping	0.28 ± 0.05
Disengagement Coping	0.13 ± 0.03
Raw Coping Scores <sup>a</sup>	
PCE Coping	2.75 ± 0.66
SCE Coping	2.78 ± 0.54
Disengagement Coping	1.75 ± 0.66
Raw Coping Scores by Level <sup>b</sup>	
PCE Coping	
“Not at all”	2 (3)
“A little”	23 (37)
“Some”	27 (43)
“A lot”	11 (17)
SCE Coping	
“Not at all”	0 (0)
“A little”	18 (29)
“Some”	39 (62)
“A lot”	6 (10)
Disengagement Coping	
“Not at all”	30 (48)
“A little”	24 (38)
“Some”	7 (11)
“A lot”	2 (3)

Note. PCE = Primary Control Engagement; SCE = Secondary Control Engagement.

<sup>a</sup>Range = 1-4 (1 = “not at all,” 2 = “a little,” 3 = “some,” and 4 = “a lot”).

<sup>b</sup>To obtain raw coping scores by level, we rounded each individual’s average for that level to the nearest whole number (e.g., an individual’s average raw PCE score of 1.92 rounds to 2, or “a little”)

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**TABLE 3**

Multiple Regressions with Percent Non-Attendance and HRQOL as Outcome Variables

DV	IV	$\beta$	b	t	p	95% CI	R <sup>2</sup>	F
Percent Non-Attendance	Caregiver Coping	-	-	-	.23	-	0.07	1.47
	PCE Coping	-0.28	-1.38	-2.00	.05 <sup>†</sup>	[-2.77, -0.00]	-	-
	SCE Coping	0.13	0.60	0.93	.36	[-0.70, 1.91]	-	-
Youth HRQOL	Disengagement Coping	-0.08	-0.68	-0.53	.60	[-3.25, 1.89]	-	-
	Caregiver Coping	-	-	-	.06	-	0.12	2.62
	PCE Coping	0.16	57.80	1.15	.26	[-42.98, 158.57]	-	-
Engagement	SCE Coping	0.28	100.88	2.05	.05 <sup>*</sup>	[2.42, 199.34]	-	-
	Disengagement Coping	0.04	27.71	0.30	.77	[-159.31, 214.72]	-	-

\*  $p < .05$

<sup>†</sup>  $p = .05$

Note. DV = Dependent Variable; CI = Confidence Interval; HRQOL = Health-Related Quality of Life; IV = Independent Variable; PCE = Primary Control Engagement; SCE = Secondary Control Engagement

**TABLE 4**

## Short-Answer RSQ Responses: Caregiver Coping Strategies

Coping Style	Specific Coping Strategy	Sample Responses
PCE	Prevention efforts	“Ensuring she stays hydrated” “In our home we tend to eat a lot more organic”
	Managing youth’s pain/treatment	“When my kids are having a crisis, I do everything in my power to keep them comfortable!” “Making sure she takes medicine”
	Problem Solving	“Identify solutions that work and make plans that include solutions” “Always beng [sic] flexible when it comes to changing our schedule and always being prepared when traveling”
	Research	“I may search the internet on different sickel [sic] cell treatments” “Doing research on sickle cell, give [sic] current information”
SCE	Distraction/relaxation/pleasurable activities	“Make myself relax, swim, exercise to relieve stress” “Listen to music, drive, read a book”
	Religious/spiritual coping	“Pray and trust God” “Pray for the bad days to weigh out”
	Cognitive coping	“I try to handle my situations alway [sic] thinking the best no matter the difficulty” “Remembering he has no control over his illness”

Note. PCE = Primary Control Engagement; SCE = Secondary Control Engagement