

# Accepted Manuscript

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Soumitri Sil, Ph.D., Carlton Dampier, M.D., Lindsey L. Cohen, Ph.D.

PII: S1526-5900(16)30069-4

DOI: [10.1016/j.jpain.2016.05.008](https://doi.org/10.1016/j.jpain.2016.05.008)

Reference: YJPAI 3258

To appear in: *Journal of Pain*

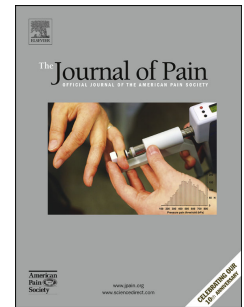
Received Date: 2 November 2015

Revised Date: 29 April 2016

Accepted Date: 24 May 2016

Please cite this article as: Sil S, Dampier C, Cohen LL, Parent and Child Catastrophizing in Pediatric Sickle Cell Disease, *Journal of Pain* (2016), doi: 10.1016/j.jpain.2016.05.008.

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Parent and Child Catastrophizing in Pediatric Sickle Cell Disease

Soumitri Sil Ph.D.<sup>1,2</sup>, Carlton Dampier, M.D.<sup>1,2</sup>, & Lindsey L. Cohen, Ph.D.<sup>2,3</sup>

1. Emory University School of Medicine, Department of Pediatrics
2. Children's Healthcare of Atlanta, Aflac Cancer and Blood Disorders Center
3. Georgia State University, Department of Psychology

**Corresponding Author:**

Soumitri Sil, Ph.D.

Emory University School of Medicine

Children's Healthcare of Atlanta

2015 Uppergate Drive, 426H

Atlanta, GA 30322

Phone: (404) 727-2712

Fax: (404) 727-4455

Email: Soumitri.Sil@emory.edu

**Disclosures:** Funding for this study was supported by Emory + Children's Pediatric Seed Grant Program to Soumitri Sil, Ph.D. The authors have no conflicts of interest to report.

## Abstract

Pain catastrophizing is poorly understood in children and adolescents with sickle cell disease (SCD) and their parents. The objectives of this study were two-fold: 1) to evaluate the interplay between parent and child pain catastrophizing and its impact on disability among youth with SCD, and 2) to evaluate whether child pain catastrophizing served as a mechanism that explained the relation between pain and functional disability within the context of varying levels of parent pain catastrophizing. One hundred youth (8-18 years old) with SCD and parents completed measures of pain characteristics (pain frequency and intensity), catastrophizing (Pain Catastrophizing Scale), and the outcome of functional disability (Functional Disability Inventory) in a cross-sectional study. Youth with low levels of catastrophizing demonstrated high levels of disability in the presence of high levels of parent catastrophizing. Additionally, child pain catastrophizing was a significant mechanism that partially explained the impact of higher pain frequency and pain intensity on greater levels of disability, but only at low levels of parent pain catastrophizing. High parent catastrophizing and incongruence between child and parent catastrophizing contributes to poorer functional outcomes in youth with SCD.

**Perspective:** Youth with SCD and parents with high catastrophic thinking about child pain or incongruent levels of catastrophizing are at increased risk for greater child disability. Clinicians treating youth with SCD should focus on targeting worried thinking about pain from patients and parents to facilitate improved function.

**Keywords:** sickle cell disease, catastrophizing, children and adolescents, parents, functional disability

Youth with sickle cell disease (SCD) and their parents report pain as the most distressing feature of the disease and the aspect that has the strongest negative impact on their health-related quality of life [10]. Approximately 23% of youth with SCD report recurrent or chronic pain and experience moderate to severe levels of functional disability [43]. There is growing evidence that pain catastrophizing plays a central role in the maintenance of pain and disability in adolescents with chronic pain, as well as central sensitization into adulthood [6, 20, 52, 55]. Catastrophizing is broadly conceptualized as an exaggerated negative “mental set” in response to experienced or anticipated pain [48]. Theoretical models suggest that those who catastrophize the perceived threat of pain are likely to focus more attention on pain thereby heightening pain, emotional distress, and pain-related fear, which may contribute to activity avoidance and subsequent disability. Consistent with these theories, catastrophizing appears to be one of the key mechanisms that explains the impact of pain on disability among youth with various chronic pain conditions (e.g., functional abdominal pain, musculoskeletal pain ) [7, 20, 24, 44, 48]. Furthermore, the development and maintenance of catastrophizing may involve social or relational factors in an effort to cope with the stress of the pain experience, such as soliciting parental interaction or empathic responses, which can further trigger or reinforce exaggerated pain expression and contribute to heightened pain experience [48]. To date, the role of pain catastrophizing is poorly understood in children and adolescents with SCD and their parents; it is unclear if these negative cognitions are relevant risk factors that are generalizable to pediatric SCD given the presence of both acute and chronic pain.

Relative to other chronic pain conditions (e.g., rheumatoid arthritis, musculoskeletal pain) and healthy controls, adults with SCD have been found to have higher levels of pain catastrophizing that were significantly associated with lower quality of life and higher severity of

depression [3, 22]. In children and adolescents with SCD, patients with high frequency or chronic pain were found to report moderate levels of pain catastrophizing [43] comparable to other pediatric chronic pain conditions (such as headache, neuropathic, or musculoskeletal pain) [41], which has been linked to increased emotional distress [14]. Although preliminary, these data suggest that pain catastrophizing may be a relevant risk factor among youth with SCD that persists into adulthood; however, it remains unclear whether pain catastrophizing serves as a driving force through which pain impacts disability and how parents' beliefs and reactions to pain may affect youths' pain outcomes.

Parental beliefs and reaction to pain play a key role in the onset and maintenance of pain and should be considered within the context of parent-child and family variables [39]. Emerging evidence from other pediatric chronic pain conditions suggests that parents' catastrophizing about child pain contributes to higher child-reported pain intensity, functional disability, and more school absences as well as parents' own engagement in pain-promoting behaviors [19, 32, 33, 47]. Youth with chronic pain and their parents typically demonstrate comparable levels of catastrophizing; however, incongruent levels of catastrophizing between parent and child have been linked to poorer outcomes [33]. In youth with SCD, poor parental coping and increased parenting stress were found to negatively impact child adjustment and frequency of healthcare use [14, 30], but the interaction between parent and child pain beliefs, such as catastrophizing, in SCD remains unclear. The purpose of this study was to understand the role of pain catastrophizing in youth with SCD and evaluate how parental pain catastrophizing about child pain can influence child functional outcomes.

Specifically, the primary aim of this study was to evaluate the interplay between parent and child pain catastrophizing and its impact on disability among youth with SCD. We expected

that parent catastrophizing would have a moderating impact on child catastrophizing and functional disability, such that children with low levels of catastrophizing would be most adversely affected by higher levels of parent catastrophizing. The secondary aim was to evaluate whether child pain catastrophizing serves as a mediator that explains the relation between pain and functional disability within the context of varying levels of parent pain catastrophizing (i.e., moderated mediation, see Figure 1).

## Methods

### *Recruitment*

Participants were children and adolescents and their parent presenting to outpatient comprehensive sickle cell clinics at three campus locations of a Children's Hospital in the southeastern U.S. between March 2014 and March 2015. Children and adolescents aged 8 to 18 years were eligible to participate if they were diagnosed with SCD. Recruitment primarily targeted youth who experienced disease-related pain in the past 30 days. A subset of patients with SCD who did not report any disease-related pain in the past 30 days also was recruited concurrently to encompass a full range of pain frequency. Exclusion criteria included patients or parents with significant documented cognitive or developmental disabilities and non-English speaking families. Based on medium effect sizes determined from prior studies of parent catastrophizing and response to youth with chronic pain [20, 29], a minimum of 62 patient-parent dyads were needed to achieve power of at least .80 and detect a parent-child interaction effect.

### *Study Procedures*

Institutional Review Board approval was obtained prior to study initiation. Study coordinators reviewed clinic appointment lists and collaborated with the hematologists and/or nurse practitioners to identify potentially eligible patients. Patients and their parents were introduced to the study by a trained research coordinator who assessed their eligibility. If

patients were eligible and expressed interest, the coordinator explained the study in greater detail. Parents provided written informed consent and children and adolescents provided written assent. Patients and parents were provided the option to complete electronic tablet-based or pencil-and-paper measures during a routine outpatient clinic visit while waiting for their provider. Patients and parents received a monetary incentive for the time dedicated to study participation.

### *Measures*

Background and Demographics. Detailed demographic and background information including patient and parent age, race, sex, highest education level, and annual family income were collected from caregivers. Common SCD treatments, specifically hydroxyurea and transfusions, were determined based on medical chart review.

Pain Intensity and Frequency. Children and adolescents reported on their average pain intensity over the last two weeks using a numeric rating scale (NRS), with “0” indicating no pain, and “10” reflecting worst possible pain. Patients also reported their pain frequency within the past month and duration based on items adapted from prior research [56]. Patients reported on the number of days they had disease-related pain in the past month (0-31 days), and how long (i.e., duration) they have experienced the current level of pain frequency to characterize pain chronicity. These scales have been validated in pediatric pain samples and are recommended for use in clinical studies of pain in school-age children [37].

Functional Disability. The Functional Disability Inventory (FDI) is a well-validated 15-item self-report instrument that assesses children’s and adolescents’ perceived difficulty to perform daily activities in home, school, recreational, and social settings [54]. Participants rated how much difficulty they have performing each of the activities on a 5-point Likert scale (0 = no

trouble to 4 = impossible). Total scores range from 0 to 60, with the following established clinical reference points: 0-12 indicating no/minimal disability, 13-29 indicating moderate disability, and 30-60 indicating severe disability [27]. The FDI has been found to have high internal consistency, moderate to high test-retest reliability, and good predictive validity [4, 54]. Internal reliability for the current sample was 0.91.

**Pain Catastrophizing.** The Pain Catastrophizing Scale, child (PCS-C) and parent (PCS-P) reports, are both 13-item questionnaires that assess thoughts and feelings about pain [6, 17]. Children completed the PCS-C regarding catastrophic thinking related to their own pain, whereas parents completed the PCS-P to assess parental pain catastrophizing about their child's pain. Items are rated on a 5-point Likert scale (0 = mildly to 4 = extremely). Total scores range from 0 to 52, with higher scores reflective of greater catastrophic thinking about pain. Total scores were used for analyses with the following clinical reference points for the PCS-C: low (0-14), moderate (15-25), and high ( $\geq 26$ ) [41]. The PCS-C and PCS-P have been used frequently in pediatric chronic pain research and well-validated in samples of youth with chronic pain and their parents [6, 41, 46]. Internal reliabilities for the current sample were 0.92 for the PCS-C and 0.90 for the PCS-P.

#### *Statistical Analyses*

All data were entered into REDCap<sup>®</sup>, a secure web application for online surveys and databases, and then exported into SPSS version 21 for analyses. There were no missing data for pain characteristics, parent and child reports of pain catastrophizing, or functional disability. Descriptive data were computed on demographics variables and measures of pain, functional disability, and pain catastrophizing to determine whether the data met the underlying assumptions of the proposed analytic procedures (e.g., skewness). Bivariate correlations were



conducted to identify potential covariates and examine the relation among pain intensity and frequency, functional disability, and child and parent pain catastrophizing.

Next, to test the moderating role of parent catastrophizing on the relation between child catastrophizing and functional disability, predictor variables were centered to aid in interpretation. Hierarchical multiple regression analysis was conducted and the interaction effect was probed using the data analytic techniques described by Hayes [21] and Preacher, Rucker, and Hayes [42].

Finally, we constructed a moderated mediation model in which pain intensity and pain frequency were linked directly with functional disability and indirectly through child pain catastrophizing, taking into account parent catastrophizing as a moderator between child catastrophizing and disability (Figure 1) [42]. Testing for significant moderated mediation involved examining whether the joint product ( $\alpha\beta$ ) of the regression coefficients relating the independent variable to the mediating variable (e.g., pain frequency  $\rightarrow$  child catastrophizing; coefficient  $\alpha$ ) and the mediating variable to the dependent variable (i.e., child catastrophizing  $\rightarrow$  functional disability; coefficient  $\beta$ ) differed significantly from zero at different levels of the moderator (i.e., low, mean, and high levels of parent catastrophizing). Specifically, 5,000 bootstrap samples were analyzed to produce a bias-corrected 95% confidence interval (CI) for possible joint product ( $\alpha\beta$ ) coefficient values based on an empirical sampling distribution [50]. A joint product ( $\alpha\beta$ ) coefficient 95% CI that did not contain zero indicated significant mediation. In addition, significant mediation in the presence of a nonsignificant direct path from pain to functional disability ( $c'$ ) indicated full mediation, whereas significant mediation in the presence of a significant direct path indicated partial mediation [34]. This method of moderated mediation

testing was utilized as it is the most robust way to control Type I and Type II errors [35, 36] compared to other methods, such as Baron and Kenny [2] that tend to inflate either or both.

## Results

### *Sample Demographic Characteristics*

Only 7 families approached for enrollment (4%) refused to participate due to lack of interest (n=3), time constraints (n=2), legal guardian was unavailable (n=1), or was too tired (n=1). An additional 3 patients (1.6%) who were approached for enrollment were ineligible due to significant cognitive impairment that would have interfered with completion of study measures. The sample included 100 children and adolescents with SCD and their parents seen in comprehensive sickle cell clinics across three children's hospital campuses. Patients were on average 13.54 years old ( $SD = 2.7$ ), and the majority of patients were female (61%), Black or African American (94%), and had hemoglobin type HbSS (77%). Caregivers were primarily mothers (86%), Black or African American (93%), and married (42%). Caregivers reported having an average of 1.13 ( $SD = 0.81$ , range 0-4) family members (not including the patient) with a history SCD (Table 1). There were no significant differences in patient or parent demographics by campus.

### *Patient Clinical Characteristics*

Descriptive statistics on pain characteristics, catastrophizing, and disability revealed skewness within the limits of normal distribution [49]. No significant differences in clinical characteristics were found based on campus location. Consistent with prior studies of youth with SCD, patients reported moderate average pain intensity ( $M=4.15$ ,  $SD=2.8$ , range = 0-10) with an average of 11 days ( $SD=10.08$ , range 0-31) of pain in the past month (Tables 1-2) [9, 13, 53]. Of the full sample, n=20 reported no disease-related pain in the past month. Approximately 40% of patients reported acute pain over the past month (i.e., between 1 and 11 days), and 40% of

patients reported chronic pain (i.e., pain on a majority ( $\geq 12$ ) of days in the past month that persisted for an average 1.2 years ( $SD = 1.95$ ) (see [43] for additional details on defining acute and chronic SCD pain). Of the total sample, 60% were prescribed hydroxyurea, of which 11% ( $n=11$ ) reported no SCD pain, 26% ( $n=26$ ) reported acute pain, and 23% ( $n=23$ ) reported chronic pain. Only 16% of patients were on chronic transfusions, of which  $n=7$  reported no SCD pain,  $n=6$  reported acute pain, and  $n=3$  reported chronic pain based on pain frequency in the past month.

The sample was characterized by moderate levels of functional disability ( $M = 15.56$ ,  $SD = 11.52$ , range = 0-42) [27] and moderate to high levels of pain catastrophizing reported by youth ( $M = 25.25$ ,  $SD = 12.21$ , range = 0-52) and parents ( $M = 26.76$ ,  $SD = 11.09$ , range = 0-51) [41] (Table 2). Greater levels of functional disability were significantly related to higher pain intensity, higher pain frequency, and greater parent and child pain catastrophizing (all  $p$ 's  $< .01$ ) (Table 2). Patient sex, parent reporter (mother vs. father), parent education, parent marital status, annual family income, and family history of SCD were not significantly associated with outcome variables. Independent samples  $t$ -tests revealed no significant difference in pain characteristics or outcomes of interest based on patient's treatment with hydroxyurea. However, patients on chronic transfusions reported significantly fewer pain days in the past month ( $M=4.44$ ,  $SD=4.82$  vs.  $M=12.26$ ,  $SD=10.35$ ), lower pain intensity ( $M=2.81$ ,  $SD=2.66$  vs.  $M=4.4$ ,  $SD=2.77$ ), and lower functional disability ( $M=8.38$ ,  $SD=7.77$  vs.  $M=16.92$ ,  $SD=8.38$ ) than patients not receiving transfusions ( $t$ 's ranging from -1.67 to 0.07, all  $p$ 's  $< .05$ ). Similar to other samples of youth with SCD, child age was significantly related to functional disability, such that older children had greater disability ( $r=.20$ ,  $p < .05$ ) [8, 25]; therefore, child age and chronic transfusion status were controlled for in primary analyses.

*Moderation Analysis*

A hierarchical multiple regression analysis examined the extent to which parental pain catastrophizing moderated the relation between child pain catastrophizing and child functional disability, controlling for child age, transfusion status, pain intensity, and pain frequency. The overall model was significant,  $F(7, 92)=4.61, p < .001$ , explaining 26% of the variance in disability (Table 3). As predicted, a significant two-way interaction emerged between parent pain catastrophizing and child pain catastrophizing in predicting disability. To examine the nature of the interaction effect, regression lines were plotted for patients with high (+1 SD) and low (-1 SD) levels of parent catastrophizing (Figure 2). For parents who reported lower levels of catastrophizing, higher levels of child catastrophizing were associated with higher levels of disability. In contrast, for parents reporting higher levels of catastrophizing, child pain catastrophizing was not predictive of disability; that is, regardless of their own level of catastrophizing, youth demonstrated moderate levels of disability in the presence of high parental catastrophizing.

*Moderated Mediation Analysis*

We tested a moderated mediation path analysis (Figure 1) consisting of the direct and indirect associations among pain intensity, pain frequency, child catastrophizing, and functional disability taking into account different levels of parent catastrophizing (Table 4). After controlling for child age and transfusion status, results showed that child pain catastrophizing was a significant mediator or mechanism that partially explained the impact of higher pain frequency on greater levels of disability ( $c' = 0.21, p = .07$ ), but only at low levels of parent pain catastrophizing ( $\alpha\beta = 0.10, 95\% \text{ CI} = [0.02-0.24]$ ). At high levels of parent catastrophizing, the

joint product 95% CI for child catastrophizing ( $\alpha\beta = 0.004$ , 95% CI = [-0.08-0.09]) contained zero, indicating no significant mediation at high levels of parent catastrophizing.

Child catastrophizing also was a significant mediator that explained the impact of higher pain intensity and greater levels of disability ( $c' = 0.68$ ,  $p = .09$ ), at low levels ( $\alpha\beta = 0.32$ , 95% CI = [0.06-0.82]) and mean levels ( $\alpha\beta = 0.16$ , 95% CI = [0.003-0.48]) of parent pain catastrophizing. At high levels of parent catastrophizing, the joint product 95% CI for child catastrophizing ( $\alpha\beta = 0.01$ , 95% CI = [-0.25-0.30]) contained zero, indicating no significant mediation.

### Discussion

To date, the influence of pain catastrophizing within the context of SCD is unclear, despite evidence among other chronic pain conditions that suggests catastrophizing is a key component that contributes to the pain experience. Our findings extend the literature on pain catastrophizing in pediatric SCD and suggest that youth with SCD and their parents demonstrate moderate to high levels of catastrophizing about child pain, which is comparable to levels of catastrophizing found among other pediatric chronic pain conditions [20, 33, 41, 52]. To our knowledge, this is the first study to begin examining the complexity between parent and child catastrophizing of child pain and the important role they play in understanding pain and functional disability among youth with SCD.

Results from this study suggest that children with low catastrophizing are adversely affected in their functional disability by high parent catastrophizing. That is, youth with low catastrophizing demonstrated moderate levels of disability in the presence of high parent catastrophizing, which is comparable to the level of disability reported by youth with high catastrophizing. Additionally, youth with high catastrophizing demonstrated greater disability in

the presence of low parent catastrophizing. Thus, high parent catastrophizing and incongruence between catastrophizing (low child – high parent; high child – low parent) resulted in poorer functional outcomes. Consistent with theoretical models of catastrophizing and pain, parental catastrophizing about child pain may serve to elicit or trigger more pain behaviors or worries from youth, further exacerbating parents' own worries and inadvertently reinforcing heightened pain expression and suffering [33]. Although discrepancies between parent and child report is more likely for internal experiences, such as catastrophizing [5], incongruence between parent-child catastrophizing may indicate conflicting strategies to manage pain, invalidation of perspective, poor communication, or broader issues related to problematic family functioning. These discrepancies between parent-child dyads are important considerations that likely impact the family's ability to effectively manage pain at home.

A secondary objective of this study examined whether the impact of pain on disability was better explained through child catastrophizing within the context of varying levels of parent catastrophizing. Results suggest that child catastrophizing was at least one mechanism that explained the impact of pain frequency and pain intensity on disability, but only in the presence of low parent catastrophizing about child pain. This may suggest that when parental interactions and communications are not worried or distressed, that a child's own level of catastrophizing has the opportunity to play a salient role in explaining the negative impact of pain on functional disability. In contrast, within the context of high parent catastrophizing, other family factors such as family functioning or parenting behaviors may have a stronger and subsuming role than the child's own level of catastrophizing. This is consistent with Palermo and Chambers' [39] integrative model of parent and family factors in pediatric chronic pain. That is, parent variables such as parental reinforcement or solicitousness through catastrophic thinking, are viewed within

a broader context of parent-child interaction, which is further rooted within the larger familial environment and functioning. For example, among youth with other recurrent pain conditions (e.g., migraine headaches), increased pain was associated with greater functional disability among youth from more disrupted family environments but not adaptive family environments [31]. The complex relationships among family, parent, and parent-child factors and child pain necessitate multilevel assessment and offers opportunities to intervene at the individual, dyadic, and family levels.

Notably, the role of catastrophizing must be viewed within the context of the complexity of SCD. Although many comparisons can be drawn between SCD and other chronic pain conditions, such as pain onset during childhood or adolescence and negative consequences on physical, emotional, and social domains of functioning, SCD remains a genetic and life-limiting disease [8, 25, 40]. As such, pain in SCD may be associated with disease-related medical complications, in which some level of catastrophizing may facilitate appropriate and needed medical care. Similar to other pain conditions, child catastrophizing may be one strategy to communicate verbal and nonverbal distress to others about pain and related SCD symptoms [52]. However, the interpretation of pain catastrophizing as a form of communication requires a level of empathy from parents that may be influenced and further exacerbated by parent's own catastrophic thinking about child pain [16, 18]. Additionally, child suffering related to pain elicits significant parent distress and concern that may be linked to parent struggles in gaining control and mastery over child pain [26]. Considerable parenting stress is known to predict greater healthcare use among youth with SCD and negatively impact child quality of life [1, 30, 38]. Therefore, it will be important to better understand how parental pain catastrophizing is related to parental behaviors in response to pain, particularly within the context of racial and

cultural influences [12]. For example, 37% of participants lived in a single parent household, in which a nonmarital coparent may be actively involved in childrearing and contribute to the larger family environment and functioning [15]. Thus, the overall impact of catastrophizing likely varies based on both intrapersonal and interpersonal characteristics between parent and child as well as other family members, suggesting the need to consider the role of parenting behaviors and response to pain in conjunction with catastrophic thinking and cultural factors to expand our understanding of the effects on child pain outcomes.

The interpretation of study findings should be considered within the context of a few limitations. First, the cross-sectional study design limits the ability to draw any causal inferences of the role of catastrophizing as a driving force that explains the impact of pain on disability. Results from this study will require additional study with a longitudinal design to test a predictive, causal model. Second, parent catastrophizing was primarily reflective of maternal catastrophizing about child pain; thus, the similarities or differences in catastrophizing by fathers or other family members in the home and the contributions it plays on child beliefs and disability remains unknown. Future examination of family functioning and factors contributing to high catastrophizing among both parents (e.g., parent history of SCD or other chronic pain conditions) of youth with more frequent pain is needed to clarify the interaction of parent and family factors and their influence on long-term functioning. Lastly, the sample primarily targeted patients with acute and chronic pain frequency, which was determined based on patient self-report and is subject to bias and random measurement error [23]. A more standardized assessment of pain frequency and a larger sample that is more representative of the clinic population may be warranted in future research to improve generalizability and statistical power.



These limitations notwithstanding, the generalizability of the study findings offer important clinical implications for pain assessment and treatment among youth with SCD. Clinicians and researchers interested in the prevention and treatment of recurrent and chronic pain in SCD should consider routine assessment of both child and parent pain beliefs, such as catastrophic thinking about pain. Patients and/or parents presenting with high levels of worried thinking about pain that may predispose patients to poor functional outcomes will benefit from education and training on how to manage and alter these beliefs. For example, current psychological treatments for chronic pain, such as cognitive-behavioral therapy, have been found to be effective in improving daily functioning and reducing pain intensity by reducing catastrophic thinking and beliefs about pain and improving pain coping self-efficacy [11, 28, 45, 51]. Therefore, early identification of these patterns in catastrophic thinking may help facilitate engagement and referrals for behavioral pain treatments designed to target maladaptive thoughts about pain and improve function over time.

### **Acknowledgements**

We would like to thank the patients and families for their time and participation in making this study a success. We would also like to thank study coordinators Leann Schilling, Shelley Mayes, Natasha Morris, Mitchell Turner, Anne Felder, and Amanda Watt. This study was funded by the Emory + Children's Pediatric Seed Grant Program to Soumitri Sil, Ph.D.

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## Figure Legends

Figure 1. Illustration of a moderated mediation model. Independent variables (IVs) affect dependent variable (DV) indirectly through mediator (M); the strength of the indirect effect (mediator) is contingent on the level of the moderator (V).

Figure 2. Interaction effect with parent catastrophizing as a moderator of child catastrophizing and functional disability. Estimated regression lines showing predicted functional disability scores for patients with low (-1 SD) and high (+1 SD) levels of parent catastrophizing. Graphed lines were plotted by selecting values one standard deviation above and below the mean for parent catastrophizing and child catastrophizing; these values were multiplied by their unstandardized regression coefficients to obtain values for plotting the predicted regression lines.

## Table Legends

Table 1. Sample characteristics (n=100)

Table 2. Descriptive statistics and correlations for pain characteristics, catastrophizing, and functional disability

Table 3. Multiple regression analysis examining the moderating effect of parent catastrophizing on the relation between child catastrophizing and child functional disability.

Table 4. Child pain catastrophizing as a mediator (M) of pain frequency and pain intensity (IVs) and functional disability (DV), at values of  $\pm 1$  SD of parent pain catastrophizing as a moderator (V), controlling for child age and transfusion status.

Table 1. Sample characteristics (n=100)

Child Characteristics		
	<u>M (SD)</u>	<u>Range</u>
Child Age	13.54 (2.74)	8-18
	<u>N (%)</u>	
Child Sex (female)	61	
Hemoglobin type		
HbSS	77	
HbSC	15	
HbS <sup>+</sup> Thal	5	
HbS <sup>0</sup> Thal	3	
Pain Frequency in past month		
0 days	20	
1-3 days	6	
4-10 days	34	
12-16 days	19	
20-28 days	7	
30-31 days	14	
SCD Treatments		
Hydroxyurea	60	
Chronic Transfusions	16	
Parent/Caregiver Characteristics		
	<u>M (SD)</u>	<u>Range</u>
Caregiver Age	41.82 (6.54)	27.64-62.79
	<u>N (%)</u>	
Parent/Caregiver reporter		
Mother/stepmother	86	
Father/stepfather	13	
Grandmother	1	
Marital status		
Married	42	

Single	37
Divorced/Separated	17
Widowed	4
Parent race and ethnicity	
Black or African American	93
Biracial/Multi-racial	2
Hispanic	1
American Indian/Alaskan Native	1
Prefer not to Answer	3
Number of Family Members with SCD	
0	27
1	50
2	18
3	4
4	1
Highest grade completed by parent	
High School or less	28
Some college	30
College degree	26
Graduate or Professional degree	16
Annual family income	
≤\$10,000	22
\$10,001-20,000	16
\$20,001-30,000	8
\$30,001-50,000	22
\$50,001-75,000	12
≥\$75,001	12
Prefer not to answer	8

Table 2. Descriptive statistics and correlations for pain characteristics, catastrophizing, and functional disability

	1	2	3	4	5	Mean	SD
1. Pain Intensity (0-10)	-					4.15	2.80
2. Pain Frequency (0-30)	.43**	-				11.01	10.08
3. Child Catastrophizing (0-52)	.26*	.28*	-			25.25	12.21
4. Parent Catastrophizing (0-52)	.29*	.21*	.41**	-		26.76	11.09
5. Functional Disability (0-60)	.31*	.35**	.31*	.30**	-	15.55	11.52

\*  $p < .01$ , \*\*  $p < .001$  (possible range of scores)

Table 3. Multiple regression analysis examining the moderating effect of parent catastrophizing on the relation between child catastrophizing and child functional disability.

Variable Name	$\beta$	SE	$p$	$R^2$
Child Age	0.38	0.41	ns	
Transfusion Status	-2.70	3.09	.07	
Pain Intensity	0.50	0.42	.07	
Pain Frequency	0.16	0.12	ns	.19
Child Catastrophizing	0.16	0.10	.07	
Parent Catastrophizing	0.12	0.11	ns	.23
Child Catastrophizing X Parent Catastrophizing	-0.02	0.01	<.05	.26

Table 4. Child pain catastrophizing as a mediator (M) of pain frequency and pain intensity (IVs) and functional disability (DV), at values of +/- 1 SD of parent pain catastrophizing as a moderator (V), controlling for child age and transfusion status.

IVs	Values of V (Moderator)	IV on M (Path a)	M on DV (Path b)	Conditional Indirect Effect (a x b)	Direct Effect (Path c')
		Est. (SE)	Est. (SE)	Est. (95% CI)	Est. (SE)
Pain Frequency	Low Parent Catastrophizing	0.29 (0.13)*	0.17 (0.10)*	0.10 (0.02, 0.25)*	0.21 (0.12) <sup>†</sup>
	Mean Parent Catastrophizing			0.05 (-0.002, 0.15)	
	High Parent Catastrophizing			-0.001 (-0.08, 0.09)	
Pain Intensity	Low Parent Catastrophizing	0.93 (0.43)	0.17 (0.10)*	0.32 (0.06, 0.82)*	0.68 (0.40)
	Mean Parent Catastrophizing			0.16 (0.003, 0.48)*	
	High Parent Catastrophizing			0.01 (-0.25, 0.30)	

\*  $p < .05$ , <sup>†</sup>  $p = .07$

Note: Values of parent catastrophizing are the mean +/- 1 SD from the mean. Results are based on 5000 bootstrap samples.



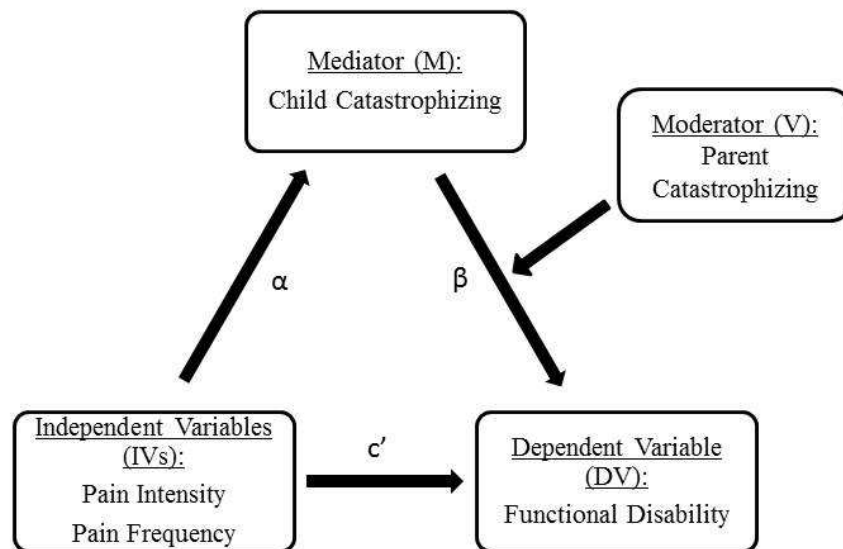


Figure 1. Illustration of a moderated mediation model. Independent variables (IVs) affect dependent variable (DV) indirectly through mediator (M); the strength of the indirect effect (mediator) is contingent on the level of the moderator (V).

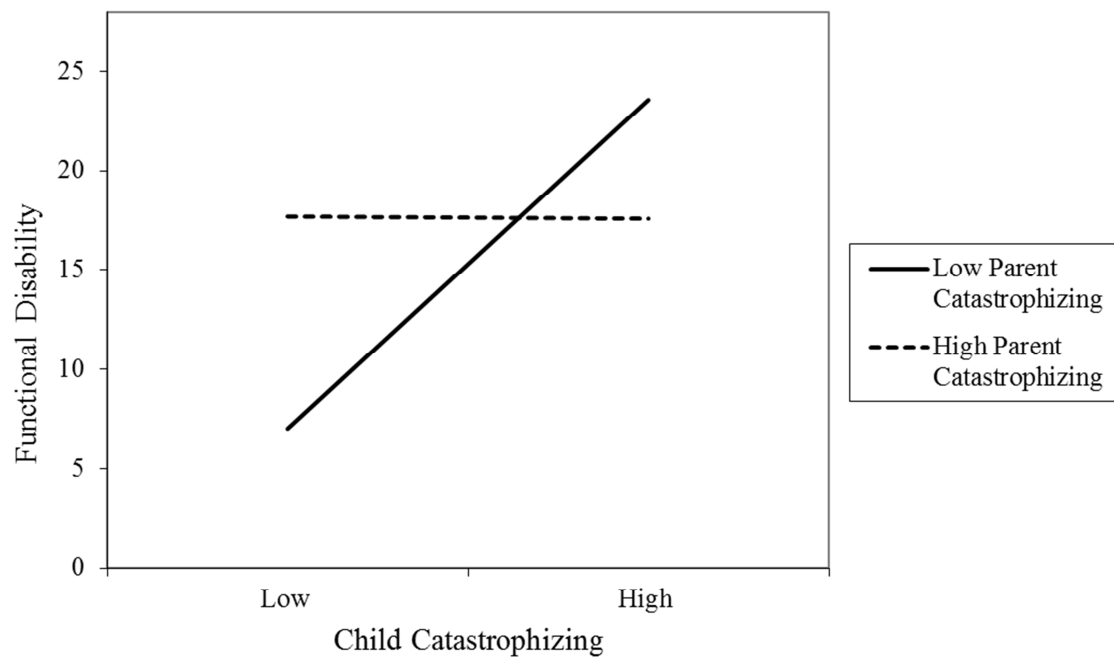


Figure 2. Interaction effect with parent catastrophizing as a moderator of child catastrophizing and functional disability. Estimated regression lines showing predicted functional disability scores for patients with low (-1 SD) and high (+1 SD) levels of parent catastrophizing. Graphed lines were plotted by selecting values one standard deviation above and below the mean for parent catastrophizing and child catastrophizing; these values were multiplied by their unstandardized regression coefficients to obtain values for plotting the predicted regression lines.

### Highlights

- Youth with SCD and their parents report high pain catastrophizing about child pain.
- Youth with low pain catastrophizing have higher disability when their parents have high catastrophizing.
- Child catastrophizing explained the link between high pain frequency and intensity on functional disability, only when parent catastrophizing was low.
- High parent catastrophizing and differences in parent-child catastrophizing leads to poorer functioning in youth with SCD.