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The Effect of Parental Mental Health on Proxy reports of Health-related Quality of Life in Children with Sickle Cell Disease

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Abstract

Background—The objectives of this study were to evaluate factors that influence agreement between parent-proxy and child self-report of health-related quality of life (HRQL) in sickle cell disease. We hypothesized that the mental health of the parent, parental HRQL and child characteristics would affect agreement.

Procedure—In a cross-sectional study of children with sickle cell disease, HRQL of the child and the parent's HRQL and mental health were assessed. The effect of parent and child characteristics on agreement between parent-proxy and child self-report of HRQL were determined.

Results—Rates of agreement between parent-proxy and child self-report of HRQL ranged between 42–49%. Parents with increased symptoms of distress had an increased odds of reporting a worse physical (Odds Ratio (OR) 1.12) and psychosocial HRQL (OR 1.10) compared to the child's self-report. Severe sickle cell disease was associated with an increased odds of the parent reporting the child's physical HRQL was worse, (OR 4.68) compared to the child's self-report.

Conclusions—Greater symptoms of distress in the parent are associated with worse parent-proxy report of the child's HRQL. Severe sickle cell disease is associated with greater disagreement between parent-proxy and child self-report of HRQL. These findings broaden our understanding of factors that influence proxy-reporting of a child's HRQL.

Keywords

sickle cell disease; health-related quality of life; family factors; well being

Introduction

It is well known that parent-proxy reports of health-related quality of life (HRQL) can be useful when a child is too young or too ill to self-report.[1,2] Parent-proxy reports can also provide valuable complementary information when self-reports are available. For example, a

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parent's assessment of their child's HRQL is a major determinant of the child's health care utilization.[3] However, the degree to which parent-proxy reports of HRQL agree with corresponding self-reports has been shown to vary as a function of the HRQL domain, the instrument used, the child's age and a number of additional parent and child characteristics. [1,4,5] For instance, agreement is usually better in domains of HRQL that are observable such as physical functioning[1] [6] and for children with a chronic disease compared to healthy children.[1,7] However, the direction of the differences in agreement may vary and are important to examine.[8] Although it has been shown that parents usually report worse HRQL for their child than the child self-reports, in a recent study of children with cerebral palsy those who report pain are more likely to self-report worse HRQL than their parent reports.[8] Sickle cell disease is characterized by frequent and recurrent pain events and is also associated with chronic pain due to complications such as avascular necrosis. Severe sickle cell disease may impact how well parent and child agree when reporting HRQL of the child, especially since pain is the most frequent complication of the disease. Given that older age is associated with more hospitalizations for pain and longer hospital stays,[9] age is likely a factor in parent-child agreement for HRQL. Therefore, understanding the factors that impact agreement between parent-proxy and child self-report of HRQL is essential.

Similar to other chronic diseases, prior research in children with sickle cell disease has identified that parents tend to report worse HRQL for their child than the child self-reports. [10,11] While there has been considerable research into factors associated with proxy-child agreement,[1,12] such factors have not been described within a sickle cell disease population. It is not immediately clear whether factors associated with agreement between child self-report and proxy report from different chronic disease populations immediately translate to children with sickle cell disease. As an example, studies that have linked parental stress to increased proxy disagreement considered European populations of children with cerebral palsy,[8] inflammatory bowel disease,[13] and psychiatric disorders[14] and a Canadian population of children with attention-deficit/hyperactivity disorder.[15] Furthermore, there is evidence that parents' mental health can influence or distort how they proxy report their child's well being. [16,17] Thus, because children with sickle cell disease may be too ill or too young to self-report their own HRQL, it is important to evaluate not only child characteristics that influence parents' proxy reports of the child's HRQL but also parental characteristics.

The purpose of this study was to evaluate both child and parental factors that may influence the agreement of parent-proxy and child self-reports of HRQL in children with sickle cell disease.[18] Our primary hypothesis was that parental characteristics such as decreased HRQL or worse mental health were associated with greater proxy disagreement resulting from lower parent-proxy reports of the child's HRQL. In addition, we hypothesized that older children and those with more severe disease might also display increased disagreement between parent-proxy and child-self reports.

Methods

Study Setting and Subjects

We conducted a cross sectional study of children with sickle cell disease and their parent/caregiver from January 1, 2006 through June 30, 2007 as part of a larger study. This larger study examined the psychometrics of the Pediatric Quality of Life Inventory (PedsQL™) 4.0 generic core scales and evaluated the impact that family income had on HRQL of children with sickle cell disease.[19,20] Eligible study subjects for the current study included children ages 5 to 18 years with sickle cell disease and their parents who presented to the Midwest Sickle Cell Center for a routine check-up. HRQL measures were administered and self completed in the clinic. Children ages 5–7 years old were administered the measures

with the aid of the research assistant following the instructions recommended by the guidelines of the PedsQL™ 4.0 generic core scales.[21] Demographic and medical data were obtained from the parent or child using a demographic/phenotypic form and through review of the child's medical record. The Institutional Review Board of the Children's Hospital of Wisconsin/Medical College of Wisconsin approved the study. Informed consent was obtained from the parent and assent from children 7 years of age or older.

Primary Outcome: Agreement

Agreement between parent-proxy report and child self-report of HRQL was the primary outcome. Similar to other studies, we defined agreement as the absolute value of the difference between parent-proxy and child-self reports being less than one-half of the standard deviation of the child-reports. [8,22] Disagreement was then further subdivided according to whether the parent report was less than the child report by more than one-half of the child report SD ("parent < child disagreement"), or greater than the threshold in the opposite direction ("parent > child disagreement"). The standard deviation of the child-self report was used since this was slightly larger than the parent-proxy report, implying a more conservative definition of disagreement.

Health-related Quality of Life in Children with Sickle Cell Disease

The PedsQL™ 4.0 generic core scales parent-proxy and child self report questionnaires were used to measure child HRQL. The PedsQL™ is a 23 item generic HRQL questionnaire that has a parent-proxy report for children ages 2 through 18 years and a child self-report questionnaire for children ages 5 to 18 years.[21] The questionnaire yields information on the physical, emotional, social and school functioning of the child during the previous 4 weeks. It has been extensively tested in both healthy children[4,23–26] and children with chronic disease,[7,27–32] and was validated in children with sickle cell disease by our group and others.[10,19] In addition, our laboratory published additional psychometric properties on its use in children with sickle cell disease including Cronbach's alpha.[19] Mean scores are calculated based on a 5-point response scale for each item and transformed to a 0 to 100 scale with a higher score representing better quality of life. There are 4 scale scores: physical functioning, emotional functioning, social functioning, and school functioning. In addition, the PedsQL™ yields 3 summary scores: a total scale score, a physical health summary score, and a psychosocial health summary score. The total score is comprised of the average of all items in the questionnaire. The psychosocial summary score is comprised of the average of the items in the emotional, social, and school functioning scales. The physical health summary score is comprised of the average of items in the physical functioning scale and is the same as the physical functioning scale score. Missing items were handled based on the developer's recommendation which allows a scale score to be calculated if at least 50% of the items in each scale are answered.[21]

Variables

Our primary covariates of interest were parental well being, severity of sickle cell disease, [11] the presence of other chronic conditions (medical and neurobehavioral co-morbidities), child's age, and family income. These child characteristics were chosen as we have previously shown that they affect HRQL, and this may also impact agreement.[11,31,33–35]

Parental Health-related Quality of Life and Psychological Well Being—Parental HRQL was measured using the PedsQL Family Impact Module and the parent's mental health was measured using the Global Severity Index (GSI) from the Brief Symptom Inventory (BSI).

Parent Health-related Quality of Life: PedsQL Family Impact Module—The PedsQL™ Family Impact Module[36] assesses parent functioning and consists of 6 scales: physical functioning, emotional functioning, social functioning, cognitive functioning, communication, and worry. The parent HRQL summary score is determined by averaging the 20 items that make up the physical, emotional, social and cognitive functioning scales noted above. The communication and worry scales on The PedsQL™ Family Impact Module are used to assess family functioning but these were not utilized in this study. The mean scores are calculated the same as the PedsQL™ generic core scales. The module has been validated for use in other populations of families with children who have a chronic illness and our laboratory described the psychometric properties of its' performance in children with sickle cell disease including the Cronbach's alpha. [36–38] Missing items were handled according to the developer's instructions.[36]

Parent Mental Health: Global Severity Index (GSI)—The GSI is a summary measure of general distress that is calculated as an overall index from the Brief Symptom Inventory. [39] The GSI is used as a general assessment of psychological well being and is the most sensitive measure of the parent's distress level. It reflects a combination of the number of symptoms and level of distress of the parent. The symptom scales that are assessed in the measure are: somatization, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation, and psychoticism. For a sample from the community without a clinical diagnosis of a mental illness, a higher score (range 0–100) indicates more symptoms and higher levels of distress.[39] Specifically, a standardized score > 50 is above average and suggests increased symptoms and a score 63 is diagnostic of a psychiatric diagnosis. The GSI has been used in numerous other chronic disease populations[40,41] and in sickle cell disease as a measure of caregiver distress.[42,43] This index was used as the primary summary measure of the psychological well being of parents of children with sickle cell disease.

Severity of Sickle Cell Disease—Similar to our previous work, we classified disease status a priori as mild or severe disease regardless of the child's sickle cell genotype. [11,19,20,44] Children with a history of a sickle cell related stroke, acute chest syndrome, 3 or more hospitalizations in the prior 3 years, or recurrent priapism were classified as having severe disease based on criteria used for intervention with hydroxyurea or bone marrow transplantation.[45–47] All others were classified as having mild disease. There is currently no accepted standard to determine clinical severity. Therefore, classification of severity was based on past observations for each patient, a method previously shown to differentiate these groups.[44]

Presence of Other Chronic Conditions—Parents were asked to report whether they had ever been told by a health care provider that their child had any of the following medical conditions: asthma, chronic allergies/sinus trouble, chronic orthopedic/bone/joint problems, chronic rheumatic disease, diabetes, epilepsy, or other chronic medical condition.

In addition, parents were asked to report whether they had ever been told by a health care provider that their child had any of the following neurobehavioral conditions: anxiety, attentional, or behavioral problems, depression, developmental delay or mental retardation, learning problems, or speech problems. Patients were classified as having either a medical or neurobehavioral co-morbidity if they reported one or more of the above noted conditions. Co-morbidities were verified in the medical records where available.

Family Income—The parents/primary caregivers provided their total household income on a categorical scale. For those respondents that did not provide a family income (n=11), the

median household income within their census-block group utilizing street addresses was used as a proxy for income level. Census block group was identified using the U.S. Census Bureau's American Fact Finder. Data from the 2000 Census Summary File 3 were downloaded for each of the block groups identified.

Consistent with previous research, family income was categorized into three groups (< \$20,000, \$20,000–40,000 and >\$40,000).[26] We used \$20,000 as our lowest category of family income; consistent with the weighted average 2006 poverty threshold (\$20,614) for a family of 4.[48]

Analysis

Descriptive statistics were calculated for child and parent/caregiver characteristics. Categorical variables are presented as observed frequencies and proportions while continuous variables (age, income) are presented as sample medians.

Spearman's rank, intra-class correlations (ICC) and differences in mean scores were used to compare mean parent-proxy and child self-report HRQL scores. Generally, we characterized the strength of the correlation between these measures as follows: 0.4 and below as poor to fair, 0.4–0.6 moderate, 0.6–0.8 as strong correlation.[49,50]

We examined the effect of the following factors on agreement: psychological well being of the parent (assessed using the GSI T-Score), parental HRQL, family income level, child's age, child's disease severity and presence of co-morbidities. Separate logistic regression models (equivalent to a multinomial regression[51]) were used to model covariate effects on each type of disagreement versus agreement. In building the final presented models, a stepwise selection procedure based on an alpha level of 0.10 was used for variable inclusion/exclusion from the model. In order to produce equivalent estimates to the multinomial model, if a covariate was included in the regression for one type of disagreement, it was forced into the model for the other type of disagreement. All analyses were performed using SAS v9.1.3 (The SAS Institute, Cary, NC) and the R language (R Development Core Team 2007).

Results

Parent and Child Demographics (Tables I and II)

We recruited a convenience sample of patients through the sickle cell disease clinic. A total of 119 parents and patients with sickle cell disease were approached for this study. There were 33 patients who refused to participate for various reasons such as “not interested” and “not enough time”. Twenty patients were missing a corresponding questionnaire (PedsQL child or parent report or the BSI). Therefore, a total of 66 parent-child pairs (55%) completed the PedsQL Generic Core scales and BSI, although the parents of 2 of these pairs did not complete the PedsQL Family Impact Module (Tables II and II). Approximately two-thirds of the children had severe disease. The majority of parents who completed the questionnaires were mothers. The mean GSI score for parents was 52.8 (standard deviation 11.0, range 33.0–80.0). Almost 20% of our parents had a score ≤ 63 which is lower than what has been reported in other chronic diseases.[40,52]

Agreement between parent-proxy and child self report HRQL (Table III and Figure 1)

Parents rated their child's HRQL lower in all areas except emotional functioning although these differences were statistically significant only for the school functioning scale. Parent-proxy and child self-report HRQL scores displayed significant correlations in all summary scores and domains. The correlations were moderate for all areas except physical health

where there was only a weak correlation. The ICC values at best indicate moderate correlation, with the values for physical, emotional, and social functioning representing poor correlation. Figure 1 shows the distribution of agreement of the parent-proxy and child-self reports. Rates of agreement were between 42% and 49%. The frequency of disagreement generally showed the parents have a higher frequency of disagreement and rate the children worse than they rate themselves. However, in the emotional functioning domain there was greater parent > child disagreement (30%) than the parent < child disagreement (21%).

Impact of the parent's psychological well being and severe sickle cell disease on the parent-proxy report of the child's HRQL (Table IV)

The parent's GSI score, severe sickle cell disease, and age were the only variables that met inclusion in the final stepwise multinomial logistic regression model (Table IV). The parent's GSI score was associated with agreement between parent proxy and child self report of the child's physical HRQL. When the parent's GSI score was higher, reflecting symptoms of distress, there was a greater likelihood that the parent would report their child's HRQL substantially worse than the child did in both the physical and psychosocial domains. Figure 2 shows a non-parametric estimate of the relationship between parent-proxy report of the child's HRQL and child self-report HRQL and the GSI score illustrating the direction of the differences in agreement; specifically whether parents are reporting higher or lower HRQL than the children are self-reporting.

Disease severity was associated with disagreement between parent proxy report and child self-report of the child's physical HRQL (Table IV). Severe sickle cell disease was associated with an increased odds that the parent would proxy-report the child's physical HRQL as being worse than the child self-reported. Age was not associated with agreement.

Discussion

Our study demonstrates that the mental health of parents of children with sickle cell disease and disease severity influence how parents proxy-report their child's HRQL. Parents with increased symptoms of distress report the child's HRQL lower than the child self reports. Parents with little to no levels of distress tend to report their child's HRQL higher than the child self reports. Additionally, parents of children with severe sickle cell disease tend to report their child's physical HRQL worse than the child does.

This study demonstrates the significant impact that a parent's mental health has on the proxy-report of HRQL for their child with sickle cell disease. Prior work in children with other chronic diseases have demonstrated that parental stress,[8,14] negative caregiver affect[53] and poor parental global health[54] are associated with worse parent-proxy report of the child's HRQL compared to the child's self-report of HRQL. In addition, as White-Konig et al.[8,55] have demonstrated, different factors affect agreement between parent-proxy and child self-report of HRQL and the direction of agreement varies as we demonstrated with parental distress.

Severe sickle cell disease was associated with more disagreement between parent-proxy and child self-report of physical HRQL in our study. Parents had a greater tendency to report their children's physical HRQL as worse than the child with severe sickle cell disease reported. This is different than prior research in children with other chronic diseases where agreement has been found to be better in children with cerebral palsy and juvenile idiopathic arthritis when the disease is more severe[8,56] or in children with cystic fibrosis who have more disease exacerbations.[57] Our findings may be due to the fact that children with severe disease may have adjusted to their level of functioning and thus report better HRQL than their parents.

Our study, similar to others, showed low to moderate correlation of parent-proxy and child self-report of HRQL.[8,58] The relationship we found was moderate for all domains except the emotional functioning domain where parents rated children worse than the children rated themselves.

Our study did not show any association with child age and agreement between parent-proxy and child self-report of HRQL. Prior studies have shown conflicting results on the effect of age on agreement between parent-proxy and child self-report of HRQL.[1] Earlier studies have shown agreement is worse for younger children. [59] [60] In contrast, recent studies have shown greater disagreement in older children. Yeh et al.[61] found greater disagreement between parent-proxy and child self-report of HRQL in adolescents being treated for cancer compared to younger children. In a study of healthy children, there was more agreement in the physical HRQL domain for younger children and greater agreement in psychosocial HRQL in older children.[5]

Proxy evaluation of HRQL adds complementary information to the child self-report and is helpful when providing overall care to the child. For example, it is known that parent-proxy reported HRQL of the child predicts healthcare utilization.[3,62] In addition, parents are the decision makers, often shared with the child, and they play a strong role in deciding what is best for their child. The conceptual framework on which our work and that of others is based follows a proxy-proxy perspective; that is, parents assess the child's well being from the proxy's perspective.[63] The PedsQL parent-proxy report follows the proxy-proxy perspective[64]. This perspective allows complementary information from the proxy perspective and has been shown to be a valid method to assess HRQL when the child is not able to self-report HRQL.[63]

Our study is limited by its cross sectional design which prohibited us from determining if our findings persist over time. The relatively small sample size also implies that we likely had limited power to detect more subtle effects on agreement. In addition, our sample is drawn from only one site with modest recruitment rates as a convenience sample thus potentially limiting the generalizability of our results. Further research with a larger sample from multiple sites will allow us to answer these questions. Lastly, our study is limited by the self-report of parental and child demographics and the assessment of HRQL within the clinic setting. However, it is currently unknown how contextual variables[65,66], such as the environment in which HRQL questionnaires are completed, impact responses in this population of children and families affected by sickle cell disease.

Our data confirm the importance of the parent's mental health and effect on proxy-reporting of the child's health. Although proxy-reporting of a child's HRQL may be the only method to assess HRQL in some situations, every effort should be made to ascertain a child's self-report of HRQL whenever possible. In addition, attention to the parent's psychological well being if the parent is the proxy-reporter, seems prudent. Lastly, parent-proxy reporting of HRQL is valuable in and of itself as prior research has demonstrated that parent-proxy reports of a child's HRQL predict future healthcare costs for that child.[3]

In conclusion, we have demonstrated that parental distress and severe sickle cell disease are associated with parents reporting worse HRQL for their child than the child self-reports. When utilizing proxy-reporting to determine a child's HRQL, it is important to understand the factors that affect this reporting and obtain child self-report of HRQL whenever possible. More importantly, attention to the mental health of parents of children with sickle cell disease may be important in the care of the child and further research into the benefit of family-centered care would be useful.

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Abbreviations

HRQL health-related quality of life

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FIGURE 1.
Distribution of agreement between child-self and parent-proxy reports in children with Sickle Cell Disease.

FIGURE 2. Relationship between difference of parent and child reports and Global Severity Index (GSI) T-score for HRQL Summary Scores. Black lines denote LOWESS regression curves. Gray areas represent limits of agreement defined as $\pm$ SD of child scores/2.

TABLE I

Demographic and Disease Characteristics of Children with Sickle Cell Disease

	n (%)		n (%)
Age		Mild Disease Status	23 (34.8)
5 to 7 years	13 (19.7)	Stroke [†]	1 (4.3)
8 to 12 years	21 (31.8)	Acute Chest Syndrome	0 (0.0)
13 to 18 years	32 (48.5)	Recurrent Priapism	0 (0.0)
Gender, female	34 (51.5)	Vaso-occlusive crises [†]	
Race		# of hospitalizations (0–2)	10 (43.5)
Black or African-American	64 (97.0)	# of hospitalizations (3+)	0 (0.0)
Unknown	2 (3.0)	Severe Disease Status	43 (65.2)
Medical co-morbidities, Any[†]	24 (36.4)	Stroke [†]	7 (16.3)
Asthma	19 (28.8)	Acute Chest Syndrome [†]	33 (76.7)
Chronic allergies	6 (9.1)	Recurrent Priapism [†]	1 (2.3)
Chronic orthopaedic, bone or joint problems	9 (13.6)	Vaso-occlusive crises	
Chronic rheumatic disease	1 (1.5)	# of hospitalizations (0–2)	8 (18.6)
Diabetes	1 (1.5)	# of hospitalizations (3+)	28 (65.1)
Epilepsy	3 (4.5)	Genotype	
Other chronic medical condition	1 (1.5)	Hb SS	44 (66.7)
Neuro-behavioral co-morbidities, Any[†]	35 (53.0)	Hb SC	13 (19.7)
Anxiety Problems	5 (7.6)	Hb S + thalassemia	7 (10.6)
Attentional Problems	19 (28.8)	Hb S ⁰ thalassemia	1 (1.5)
Behavioral Problems	15 (22.7)	Hb SS G (Philadelphia Disease)	1 (1.5)
Depression	6 (9.1)		
Developmental Delay or Mental Retardation	3 (4.5)		
Learning Problems	18 (27.3)		
Speech Problems	7 (10.6)		

N=66,

[†]Some data missing,[†]Perinatal stroke not related

TABLE II

Characteristics of Parent / Primary Caregiver

<i>n</i>	66		
Relationship to Child		Brief Symptom Inventory	
Mother	56 (84.8)	GSI T score > 50 but <63	27 (40.9)
Father	5 (7.6)	GSI T score ≥ 63	13 (19.7)
Other Legal Guardian	5 (7.6)	PedsQL Family Impact (n=64)	
Race		Parent HRQL Summary Score	
Black or African-American	60 (90.9)	Median (Interquartile Range)	75.0 (61.3, 91.3)
Hispanic or Latino	1 (1.5)		
Unknown	5 (7.6)		
Have Sickle Cell Disease [†]	7 (10.6)		
Education			
Some high school or less	16 (24.2)		
High school diploma	11 (16.7)		
Educational Development	4 (6.1)		
Vocational / some college	22 (33.3)		
College / Graduate degree	4 (6.1)		
Unknown	9 (13.6)		
Self -Reported Family Income			
\$20,000	21 (31.8)		
> \$20,000 but ≤ \$ 40,000	16 (24.2)		
> \$40,000	18 (27.3)		
Unknown	11 (16.7)		

[†] Some data missing

GSI: Global Severity Index, HRQL: Health-Related Quality of Life

TABLE III

Relationship between child self and parent-proxy rated child HRQL

Summary Score	Parent-reported HRQL score Mean (SD)	Child-reported HRQL score Mean (SD)	Mean Difference (SE)	Mean Absolute Difference (SD)	ICC
Total score	63.4 (18.8)	67.5 (17.7)	-4.04 (2.49)	15.49 (13.48)	0.40**
Psychosocial Health	63.8 (18.5)	66.8 (19.3)	-2.96 (2.45)	15.20 (13.09)	0.44**
Physical Health	62.8 (23.5)	68.7 (19.7)	-5.86 (3.24)	19.43 (18.60)	0.27*
Domains					
Emotional Functioning	69.9 (18.9)	66.9 (22.2)	3.06 (2.91)	17.96 (15.48)	0.35**
Social Functioning	67.6 (21.2)	74.1 (24.1)	-6.53 (3.28)	20.32 (18.33)	0.32**
School/Work Functioning (n=64)*	53.3 (22.0)	59.4 (21.7)	-6.27 (2.98)*	19.00 (15.60)	0.41**

* Missing data due to two children not in school and school domain not completed;

: Spearman's Rank Correlation; ICC: Intra-class correlation;

* p < 0.05,

** p < 0.01

Table IV

Multinomial logistic regression models explaining “parent > child” and “parent<child” disagreement* versus agreement for PedsQL Physical Health and Psychosocial Health Summary Scores

	“parent < child” disagreement	“parent > child” disagreement
Physical Health		
Variable	Odds Ratio (95% CI)	Odds Ratio (95% CI)
GSI T-Score	1.12 (1.03–1.21)	0.93 (0.86, 0.999)
Severe SCD	4.68 (1.05, 20.78)	0.32 (0.07, 1.46)
Psychosocial Health		
Variable	Odds Ratio (95% CI)	Odds Ratio (95% CI)
GSI T-Score	1.10 (1.02, 1.18)	1.01 (0.95, 1.08)
Age	1.18 (0.996, 1.39)	1.05 (0.88, 1.26)

* Parent-proxy and Child-Self reports differ by more than half a standard deviation; Bold areas highlight significant differences;

GSI: Global Severity Index from Brief Symptom Inventory; SCD: Sickle Cell Disease