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Parent-Child Discrepancies in Children with Chronic Fatigue Syndrome-Like Symptomatology

Parent-Child Discrepancies in Children with Chronic Fatigue Syndrome-Like Symptomatology

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ABSTRACT In a sample of children and adolescents with symptoms related to chronic fatigue syndrome (CFS), we characterized the relationship between parent and child ratings of symptoms as well as domains of health-related quality of life (HRQOL) relevant for the assessment of substantial reductions in functioning. Parent-child dyads (N = 147) were recruited as part of a community-based epidemiological study of myalgic encephalomyelitis (ME) and CFS in Chicago. Parents and children completed the Children's Health Questionnaire (CHQ) as well as the DePaul Pediatric Health Questionnaire (DPHQ). Results show that inter-rater reliability between parent and child responses was typically strong, however, in most domains, parents of children with CFS-like symptoms rate their child's symptoms and HRQOL as more frequent and/or severe than the children themselves. Recommendations are provided for use of parent proxy and child self-report in diagnosis and implementation of case definitions.

INTRODUCTION

The illness often referred to as pediatric myalgic encephalomyelitis (ME) or chronic fatigue syndrome (CFS) is defined by persistent or relapsing chronic fatigue, an absence of medical exclusionary diseases causing fatigue, and a substantial reduction in physical functioning, school attendance and performance, and extracurricular activities and hobbies (Jason et al., 2006; Jason et al., 2015a). Although prevalence estimates for pediatric ME and CFS is

sparse, it is estimated that over 4% of a pediatric sample experience fatigue, and at least 2.05% qualify for a CFS-like diagnosis, with adolescents having higher rates of symptoms than younger children, as well as those with Latino origin (Jordan et al., 2000). There are few community-based pediatric epidemiological studies that have utilized rigorous medical evaluations. Consequentially little is known about the health-related quality of life (HRQOL) limitations

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Research is ongoing

characteristic of children with ME and CFS-like symptoms who may qualify for a diagnosis.

Similar to adults, key symptoms of ME and CFS in children include post-exertional malaise (PEM; an abnormal exhaustion following minimal physical and/or mental effort, Jason & Evans, 2012), memory and/or concentration problems, unrefreshing sleep, and pain (Jason et al., 2006; Carruthers et al., 2003). Unique pediatric features of the illness can include rashes, a more gradual symptom onset, poor school attendance, and a shorter required duration of symptoms to meet diagnostic criteria (e.g., 3 months versus 6 months for adults; Jason et al., 2006). Although fatigue, ME and CFS symptomatology, and disability (i.e., substantial reduction in functioning) are key features of the illness required for diagnosis in case definitions (e.g., Fukuda et al., 1994; Carruthers et al., 2003; IOM, 2015), little is known about disability and HRQOL in ME and CFS-like pediatric samples.

As with many pediatric illnesses, both parent and child symptom ratings are informative for the diagnosis and understanding of ME and CFS. Child self-report and parent proxy report are of particular importance in the diagnosis processes of pediatric ME and CFS, as there is no known biomarker that can be used to make diagnoses, requiring complex case definitions (e.g., Fukuda, 1994; Reeves et al., 2005; IOM, 2015 Carruthers et al., 2003; Jason et al., 2006).

Previous research in pediatric chronic illnesses have shown that parent and child ratings can be discrepant when employing widely used instruments such as the Children's Health Questionnaire (CHQ; Landgraf, Abetz, & Ware, 1996) which assesses 12 domains of general health and well-being. Waters, Stewart-Brown, and Fitzpatrick (2003) found that in a representative sample of adolescents, including both healthy and those with illnesses, adolescents viewed their HRQOL more poorly than their parents, especially in terms of general health, bodily pain, mental health, and impact on family activities. In a comparison of siblings in which one sibling had epilepsy and the other did not, Baca et al. (2010) found that children with epilepsy tended to rate their own health and

quality of life higher than their parents, showing that parents may underestimate their children's health. One study of children diagnosed with ME and CFS used the CHQ and found that children were mostly in agreement with their parents; however, parents rated their children as having lower functioning on the self-esteem, role physical, and physical functioning domains than the children themselves (Kennedy, Underwood, & Belch, 2010).

Another reason to explore parent-child discrepancies is that historically children have been seen as unreliable in their ability to describe and understand their own symptoms; though using only proxy raters discounts the children's subjective experiences and perceptions (Eiser & Morse, 2001). Discrepancies with parents tend to be greater for older children, as well as among dyads in which the parent is experiencing depression and lower socioeconomic status (De Los Reyes & Kazdin, 2005).

Clinicians must negotiate these factors and base diagnostic decisions by incorporating both the parent and children's concerns. Parents are more likely to believe a problem warrants treatment (De Los Reyes & Kazdin, 2005) and parents generally rate their children's symptoms as worse than do the children. However, any proxy rater, including parents, may not have access to all health-related information such as internalizing problems, leading to a greater likelihood of under-endorsing such symptoms (Eiser & Morse, 2001).

The current study compared parent and children's ratings among parent-child dyads of children with CFS-like symptoms and controls, hypothesizing that children would rate their HRQOL higher than their parents, assuming proxy raters would overestimate the level of disability experienced by their children. Secondly, this article assessed differences between CFS-like and control parent-child dyads, assuming there would be larger and more frequent discrepancies in the ME and CFS-like group.

METHODS

Data was collected from a community-based epidemiological study of pediatric ME and CFS of children between the ages of 5 and 18 in the Chicagoland area. Participants who screened positive for ME and CFS-like symptoms, according to our Pediatric Screening Questionnaire, attended a follow-up medical evaluation. During the initial phone screening, parents were asked to verbally consent to answering questions about their children's health. During the subsequent medical evaluation, both parents and children were asked to provide informed consent (or assent if child is under the age of 12) by a trained and IRB approved research assistant. Both the parents and children completed additional measures online via REDCap software before or during the medical evaluation appointment (Harris et al., 2009).

Participants

Those who screened positive for CFS-like symptoms during the initial phone interview with parents totaled 147 children. Children were 55.4% female ($M_{age} = 13.44$, $SD = 2.61$) and 75.7% had parents who were married or living with a partner, 36.7% had a standard college degree and 43.5% were employed full-time. In terms of race, 60.5% of youth were white/European American, 19.7% black/African American, 2.0% American Indian or Alaska Native, 1.4% Asian/Pacific Islander, 6.1% biracial, 23.6% were Latino or Hispanic, and 10.2% responded as "other" or "prefer not to respond". Percentages do not add up to 100% because of the biracial category, and because the Latino or Hispanic category is a separate variable.

The control group included 40 parent-child dyads, where the parent on the initial telephone screen did not endorse ME or CFS-like symptoms. 57.5% of these youth were female ($M_{age} = 13.45$, $SD = 2.32$), and 80% of their parents were married or living with a partner, 40% had a standard college degree, and 62.5% were employed full-time. In terms of race, 57.7% of the youth were white/European American, 20.0% black/African American, 7.5% Asian/Pacific Islander, 7.5% biracial, 20.0% Latino/Hispanic, and 7.5% "other/prefer not to

respond" (percentages do not add up to 100%, see above). Of the 40 controls, 25% of children had allergies and/or asthma, 2.5% (2.5% is equal to 1 individual) had juvenile dermatomyositis, 2.5% had major depression, and 2.5% had anxiety.

Measures

Child Health Questionnaire

The CHQ is a commonly used measure to assess HRQOL for children and adolescents, with separate forms for the parent (CHQ-PF-50) and children (CHQ-CF-87; Landgraf, Abetz, & Ware, 1996). The CHQ-PF-50 consists of 50 items that measure the children's functioning and well-being across 12 domains. The CHQ-CF-87 consists of 87 items across 11 domains. The domains for the children and parents are not identical, so only the 9 domains that are equivalent were analyzed. The CHQ is known to have good construct validity for measuring physical and psychosocial health in children with chronic illness and has good/excellent internal reliability (Drotar et al., 2006).

For scoring, raw scores of individual items were summed and transformed to a 100-point scale, with higher scores indicating better health.

DePaul Pediatric Health Questionnaire

All parent-child dyads completed the DePaul Pediatric Health Questionnaire (DPHQ; Jason et al., 2006), a 54-item self-report assessment of multiple domains of ME and CFS symptomatology as well as demographic information. Participants rated each symptom's frequency and severity over the past 3 months on a 5-point Likert scale. The frequency and severity scores of each item were transformed to a 100-point scale by multiplying by 25. To calculate symptom domain scores, the average of the frequency and severity scores were taken (e.g., for an 8-item domain, the 16 frequency and severity scores were averages). Symptom domain scores range from 0 to 100, for which 100 indicates worse ME and CFS symptoms.

DPHQ domains represent cardinal symptoms of ME and CFS (PEM, cognitive symptoms, sleep

problems, and an individual fatigue item) were defined in Jason et al.'s (2015b) factor analysis, which yielded a four-factor solution. Thus, the sleep domain consists of 3 items, the PEM domain included 8 items, and the cognitive domain consists of 8 items. The other factor included a variety of different domains and so was not included in the current study. We also used the single item fatigue rating transformed to the 100-point scale. The DPHQ has good/excellent test-retest reliability (Jason et al., 2015a) and internal reliability (Brown, Kaplan, & Jason, 2012).

RESULTS

Intraclass correlations (ICC) assessed general parent-child interrater reliability for the 9 CHQ domains the CHQ-PF-50 and CHQ-CF-87 had in common (as in Waters et al., 2003). Similarly, paired sample *t*-tests directly compared mean differences between parent-child CHQ domain scores on these 9 domains assessed by both the parent and child.

ICC's ranged from -0.07 to 0.77 for controls and 0.54 to 0.71 for ME and CFS-like parent-child dyads (see Table 1). For both groups, the role

physical domain on the CHQ had the poorest inter-rater reliability. The greatest inter-rater reliability was seen on the fatigue item on the DPHQ for controls and sleep domain on the DPHQ for ME and CFS-like dyads. Overall, results support our hypothesis that there would be reasonable (based on Cicchetti's recommendations, 1994) parent-child interrater reliability among the ME and CFS-like group.

Paired-samples *t*-tests assessed differences in mean ratings between parents and children on the 9 comparable CHQ domains and 4 DPHQ domains. For the control group, significant differences were found on the sleep and neurocognitive domains on the DPHQ, and the behavior and general health domains on the CHQ (see Table 1). For the ME and CFS-like group, there were notable differences on the PEM domain on the DPHQ, as well as on the role physical, bodily pain, behavior, self-esteem, and family activities domains on the CHQ (see Table 1).

The ME and CFS-like group had statistically significant mean differences on 6 domains, while the control group only differed on 4, and at a lower level of significance. In addition, the

| | Control (n = 40) | | | ME and CFS-like (n = 147) | | |
|---------------------|------------------|-----------------|---------|---------------------------|------------------|---------|
| | Parent M (SD) | Child M (SD) | ICC | Parent M (SD) | Child M (SD) | ICC |
| DPHQ Domains | | | | | | |
| Sleep | 8.50 (7.59) | 12.19 (9.87)** | 0.60*** | 36.79 (18.63) | 35.91 (18.56) | 0.71*** |
| PEM | 5.56 (11.60) | 8.63 (12.12) | 0.61*** | 33.23 (22.18) | 29.46 (20.78)* | 0.67*** |
| Neurocognitive | 3.75 (5.59) | 10.67 (11.91)** | 0.23 | 31.32 (22.22) | 33.72 (22.77) | 0.71*** |
| Fatigue Item | 15.94 (17.68) | 17.19 (18.71) | 0.77*** | 54.00 (22.19) | 51.72 (24.32) | 0.70*** |
| CHQ Domains | | | | | | |
| Phys. Functioning | 99.17 (4.46) | 96.84 (6.14) | -0.08 | 83.25 (21.02) | 82.93 (17.87) | 0.67*** |
| Role Physical | 98.29 (10.68) | 98.58 (5.80) | -0.07 | 76.08 (29.91) | 83.06 (26.21)** | 0.54*** |
| Bodily Pain | 90.25 (13.49) | 88.00 (13.10) | 0.51* | 59.18 (23.39) | 65.63 (25.25)*** | 0.68*** |
| Behavior | 89.46 (8.72) | 85.37 (9.74)* | 0.32 | 68.79 (20.13) | 74.98 (13.55)*** | 0.61*** |
| Mental Health | 82.13 (12.35) | 82.38 (10.54) | 0.77*** | 65.34 (19.55) | 67.09 (16.69) | 0.60*** |
| Self-Esteem | 82.40 (20.53) | 85.18 (14.00) | 0.45* | 67.28 (23.31) | 72.80 (18.87)** | 0.62*** |
| General Health | 82.25 (14.88) | 76.29 (14.53)* | 0.59** | 63.68 (19.26) | 62.59 (18.18) | 0.60*** |
| Family Activities | 90.81 (16.62) | 89.00 (16.40) | 0.69*** | 66.88 (26.42) | 70.83 (23.33)* | 0.66*** |
| Family Cohesion | 78.46 (15.69) | 76.54 (23.12) | 0.49* | 69.41 (23.88) | 66.95 (27.93) | 0.64*** |

Table 1. Intraclass Correlations and Paired Sample *t*-tests between the Parent and Child CHQ and DPHQ Domains

Note: for significance paired *t*-tests between parent and child (denoted on child mean scores) and of ICC: **p* < .05, ***p* < .01, ****p* < .001. On DPHQ domains, higher scores indicate worse frequency/severity. On CHQ domains, higher scores indicate better health. For ICC scores: poor = 0.0 – 0.39, fair = 0.40 – 0.59, good = 0.60 – 0.74 and excellent = 0.75 – 1.0 (Cicchetti, 1994).

domains that were discrepant between the control groups were ones that are commonly discrepant between parents and children, such as the children's behavior. These results support our hypothesis that parents would rate their children's symptoms and level of disability as worse than the children themselves.

DISCUSSION

Even within the context of generally good agreement, parents and children did exhibit mean differences in CHQ and DPHQ domains that could indicate clinical significance. For example, in the ME and CFS-like group, four out of five of the CHQ domains for which parents and children means differed significantly did so with a 5-point difference on the 100-point scale, which can be interpreted as clinically and socially significant (e.g., Kurtin et al., 1994; Landgraf et al., 1996; Wake et al., 2000; Waters et al., 2003). Considering the mean differences and ICC results in our study, we recommend that clinicians consider both viewpoints when dealing with parent-child dyads that may be discrepant at a clinically meaningful level. Clinicians should explore possible reasons for differing youth and parent concerns, keeping in mind that it is common for parents to rate symptoms of HRQOL as worse than the children themselves. Contextual factors known to be related such as parent depression, socioeconomic status (De Los Reyes & Kazdin, 2005), and others that may reasonably be related such as family cohesion, should continue to be accounted for.

In the ME and CFS-like group, children rated the role physical, bodily pain, behavior, self-esteem, and family activities domains as having significantly better functioning than did their parents. Children with ME and CFS-like symptoms rated their PEM symptoms as significantly better than their parents. These findings support our hypothesis, which stated that children would rate HRQOL domains higher than their parents. The current findings align with Eiser and Morse's (2001) findings that parents tend to rate HRQOL as worse than the child themselves, especially with symptoms with more external manifestations, such as pain or behavior problems.

In contrast, for the control group, children rated the sleep and neurocognitive domains significantly worse than their parents, as well as the behavior and general health domains. These children rated their symptoms as worse than their parents on the following domains: PEM, physical functioning, bodily pain, behavior, general health, and family cohesion. A possible reason is that their parents are over-endorsing occasional problems in areas where the children do not see it as ongoing or problematic in comparison to their peers. When comparing the control group to the ME and CFS-like group, it is evident that the nature of this illness causes more discrepancies in illness perception for the ME and CFS-like group. This may be because it is more difficult for a ME and CFS-like patient to communicate symptoms as well as how those symptoms affect everyday functioning.

Especially in a health-care setting, clinicians should make an extra effort to examine both ME and CFS-like symptoms and HRQOL, since symptoms are likely to vary across the two instruments.

Limitations and Future Directions

This study had several limitations. Our data was cross-sectional, and we did not examine relationships between ME and CFS symptoms and HRQOL outcomes, however, future work will examine this. It is also of importance to collect follow-up data that can assess variability in symptom severity known to characterize relapsing and remitting symptoms. Another limitation was that the sample size of the control group was smaller than the CFS-like group. This could have contributed to the attenuation of interrater reliability and statistical power to detect mean differences.

Finally, the CHQ-PF-50 and CHQ-CF-87 are not identical measures, as the domains administered to both parents and children are made up of a different number of items, presenting limitations to direct comparisons. However, we assumed that transformation to the 0-100 scale created a comparable metric for our comparisons.

In conclusion, although parent and child ratings exhibited generally good agreement, clinically significant differences did arise. Neither rater's perspective is clearly the gold standard in the

context of ME and CFS diagnosis, and therefore continuing to obtain multiple ratings to assemble a thorough picture of the children's health is of importance.

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