Informant Discrepancy in Perceptions of Sickle Cell Disease Severity

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Objectives To evaluate whether informants (children, caregivers, and physicians) differ in their perceptions of chronic disease severity and the extent to which these differences can be explained by objective indices of disease severity, and adjustment of the caregiver. Methods Participants were 58 children and adolescents between the ages of 8 and 18 years diagnosed with sickle cell disease. Information on perceptions of disease severity, caregiver adjustment, and biological markers of disease severity was obtained at a routine clinic follow-up appointment. Results Analyses indicated significant differences in perceptions of disease severity. Psychological adjustment of the caregiver and biological indices of disease severity were significant predictors of these differences. Conclusion Implications for the association between chronic disease and adjustment are discussed.

Key words sickle cell; chronic disease; child; disease severity; multiple informant assessment; adjustment.

Early studies in pediatric psychology have indicated that children with chronic illness have a universally increased risk for developing adjustment problems compared to healthy controls or normative groups (e.g., Cadman, Boyle, Szatmari, & Offord, 1987; Morgan & Jackson, 1986; Nelms, 1989). Increasingly, researchers find that the association between chronic illness and adjustment problems in children is more complex than initially thought and is contingent upon variables that have only begun to be explored. For example, severity of disease has been found to be an inconsistent predictor of adjustment problems, with some studies finding a significant association between severity and adjustment (Counterman, Saylor, & Pai, 1995; Daltroy et al., 1992) and others finding no such relationship (Bradford, 1994; Noll et al., 1996).

One potential explanation for the equivocal findings on the association between pediatric chronic illness and adjustment is that results are contingent on informant source (Thompson & Gustafson, 1996). Several studies have found that informants such as caregivers and children can differ dramatically in their perceptions of adjustment as an outcome variable (Achenbach, McConaughy, & Howell, 1987; Overholser, Spirito, & Difilippo, 2000). Consequently, conclusions drawn on the relationship between pediatric chronic illness and adjustment can differ depending on who provided the opinion. These findings have led some authors (e.g., Holmbeck, Li, Schurman, Friedman, & Coakley, 2002) to advocate for the inclusion of multiple informants and use of multi-informant analyses. However, the role of informant on the relationship between disease and adjustment has received scant attention in the pediatric chronic illness literature.

Studies explicitly evaluating informant discrepancy in child adjustment are more generally found in the psychiatric literature. These studies converge on the finding of poor child–caregiver agreement on the report of psychological symptoms, with clinically significant symptoms reported by a caregiver often not being reported by the child (Achenbach et al., 1987; Canning, 1994; Handwerk, Larzelere, Soper, & Friman, 1999; Klein, 1991). Explanations for these informant discrepancies remain largely speculative and have rarely been studied systematically, although some studies suggest that caregiver adjustment (particularly maternal...
depression) is a salient factor related to overreporting child adjustment problems (Briggs-Gowan, Carter, & Schwab-Stone, 1996; also see Richters, 1992).

Given the variations observed in informant perceptions of adjustment and negative life events, discrepancies in perceptions of disease severity may influence the relationship between disease and adjustment as well. Indeed, perceptions of disease severity have been found to be more of a decisive factor in defining the relationship between chronic illness and risk of poor psychosocial outcome than objective disease severity markers (Boekaerts & Röder, 1999). However, few studies have evaluated informant discrepancies in perceptions of disease severity. The limited research in this area has focused on perceptions of pain and demonstrates that informants (i.e., caregivers, children, and medical staff) tend to perceive a child’s pain differently (Chambers, Reid, Craig, McGrath, & Finley, 1998; Gil et al., 2000; Manne, Jacobson, & Redd, 1992). Factors underlying these differences remain speculative, but severity ratings between child and caregiver may be influenced by the latter’s adjustment (consistent with the literature on caregiver adjustment and symptom ratings of children), and ratings between other informants and physicians may be influenced by the latter largely basing his or her perceptions on available medical indices (based on findings from studies with adults; Abbott, Dodd, & Webb, 1995). Evaluating informant discrepancy in perceptions of disease severity and the factors underlying this discrepancy may help to elucidate conflicting findings in the area of pediatric chronic illness and adjustment.

Our study goals thus were to (a) evaluate whether multiple informants (children, caregivers, and physicians) differed in their perceptions of disease severity (defined as perceptions of symptom severity, usual pain, and usual functional limitation), and (b) explore whether informant discrepancies in perceptions of disease severity are systematic by evaluating their association with objective indicators of disease and caregiver adjustment. We hypothesized that informants would differ in perceptions of disease severity, with caregivers and physicians perceiving higher disease severity relative to child ratings consistent with findings in the literature that children tend to report fewer symptoms relative to other informants. We further hypothesized that the child–caregiver discrepancy would be accounted for by the psychological adjustment of the caregiver consistent with previous literature, and that child–physician and caregiver–physician discrepancies would be accounted for by objective medical indices that physicians often use to formulate a patient’s disease severity.

This investigation is the first known study to examine multiple informant discrepancies in perceptions of disease severity and their association with psychological and medical variables. We studied children and adolescents diagnosed with sickle cell disease (SCD) because SCD is associated with high variability in disease severity, affects a relatively homogeneous subset of children, and consists of specific objective disease severity markers (hemoglobin levels and disease subtype).

**Method**

**Participants**

Participants were 58 African-American patients (33 female and 25 male) between the ages of 8 and 18 years ($M = 11.8, SD = 2.77$) who attended the outpatient sickle cell clinic at a large health sciences center in the southeastern part of the United States over the course of 1 year. Most of the caregiver participants for this sample were the biological mothers of the children ($n = 49; 84\%$), followed by biological fathers ($n = 7; 12\%$), grandmother ($n = 1; 2\%$), and aunt ($n = 1; 2\%$). We kept those having someone other than a biological mother as a caregiver in the final sample, given our focus on primary caregivers overall. The average maternal and paternal educational attainment was a high-school diploma. Children participants were typically diagnosed with the most severe cases of SCD (sickle cell anemia, $n = 45; 77\%$), followed by the less severe types of sickle cell hemoglobin C ($n = 8; 14\%$) and sickle beta thalassemia ($n = 5; 9\%$). Average age at symptom onset based on caregiver report was 0.97 years ($SD = 1.22$).

**Measures**

**Demographics.** Caregivers answered questions about educational attainment and current occupation, children’s age at symptom onset, and medications the child was currently receiving.

**Objective Ratings of Disease Severity.** The medical charts of participating children were reviewed for sickle cell subtype, hemoglobin levels over the last three consecutive clinic visits, and frequency of emergency room (ER) visits during the past 12 months. Low hemoglobin levels, sickle cell anemia disease subtype, and frequent ER utilization are typically interpreted as representing more severe disease. Although these variables do not exhaust objective indicators of SCD severity and are albeit imperfect indicators, they are often linked with biological events such ischemia and neurological infarcts (Lemanek, Ranalli, Green, Biega, & Lupia, 2003).
**Caregiver Adjustment**

*Caregiver psychological symptoms.* The Symptom Checklist-90-Revised Scale (SCL-90-R; Derogatis, 1983) was used to evaluate caregivers' level of psychological distress. The SCL-90-R is a self-report measure in which respondents rate how much a given problem has distressed them in the past week using a 0 (not at all) to 4 (extremely) scale. The SCL-90-R measures multiple domains of psychological adjustment and has been found to have good reliability, validity, and cultural sensitivity (Thompson, Gil, Burback, Keith, & Kinney, 1993). We used the global symptom index from the SCL-90-R as an index of caregiver adjustment.

**Perceptions of Disease Severity**

A brief questionnaire was developed to assess perceptions of disease severity. The child version of the questionnaire asked children to rate their symptom severity, functional limitation, and pain level using a 100 mm visual analog scale with “not at all” and “worst possible” as the anchor points (e.g., “In general, how bad or severe do you think your sickle cell disease is?”). Similarly, the caregiver version of the questionnaire asked caregivers to rate their child's disease severity, functional limitation, and pain level using a 100 mm visual analog scale with “not at all” and “worst possible” as the anchor points. Attending physicians also were asked to rate the child's symptom severity and functional limitation using an identical metric.

**Procedure**

Families were approached during a routine medical follow-up appointment at an outpatient pediatric sickle cell clinic and asked to participate in a study evaluating the effect of SCD on children's social adjustment. Prospective participants were informed that the study required approximately 1 hr of their time during which both the caregiver and child would complete a set of questionnaires. Families also were told that they would receive $10 for their participation. Using forms approved by the Institutional Review Board at the health sciences center, researchers obtained informed consent from interested caregivers and assent from the children. Five families refused participation in this investigation due to time constraints. Participating caregivers and children were asked to complete the set of questionnaires independently in the examination room. The attending physician completed a measure describing his or her perceptions of the child's disease severity.

**Results**

**Informant Discrepancy on Subjective Disease Severity Variables**

Table I presents descriptive statistics (means and standard deviations) and zero-order correlations for the disease severity, demographic, and adjustment measures. Distributions of the variables were found to be relatively normal. Missing data were restricted to sample characteristic variables (i.e., reported age at symptom onset, socioeconomic status) and were estimated through sample mean substitution, which may have restricted the variance on these variables to some extent but sample size precluded use of reliable maximum likelihood estimates. Differences among informants on perceptions of disease parameters (pain, functional limitation, and symptom severity) were assessed via repeated-measures analyses of variance (ANOVA) using the General Linear Model.

Analyses indicated that child and caregiver perceptions of disease severity were statistically discrepant for perceived symptom severity, $F(1, 57) = 2.47, p = .01$, partial $\eta^2 = .10$. The direction of the discrepancy indicated that children on average perceived less symptom severity than did their caregiver. Children also were found to perceive less symptom severity ($M = 40.74$) relative to their physicians ($M = 49.68$), $F(1, 57) = 2.94, p = .09$, partial $\eta^2 = .05$, though the analysis was not statistically significant. No significant discrepancies in perceptions of functional limitation or pain were observed between informants, all $F$s < 1.00.

**Predictors of Informant Discrepancy**

We regressed original metric informant difference scores onto the predictor variables of interest (child data were subtracted from that of each of the other informants for analyses involving child discrepancy, and physician data were subtracted from caregiver data for the analyses involving only these two informants). Child age, gender, and reported age at symptom onset (per caregiver report) were entered first to control for effects of these variables on discrepancies as identified in previous research (Holmbeck et al., 2002). Subsequently, we entered caregiver adjustment (predicting child-caregiver and caregiver–physician discrepancy) or objective medical variables (predicting child–physician and caregiver–physician discrepancy) on the next step of the model. Direction of relationship was determined by the sign of regression coefficients and examination of plots.

The control variables were not significant when entered in the first step of models predicting caregiver–physician and child-physician discrepancies. However, the control variables together accounted for a significant
portion of the variance in child–caregiver discrepancies in reports of symptom severity, $R^2 = .14$, $F(3, 54) = 4.05$, $p = .04$, with reported age at symptom onset being the only significant unique predictor in this step of the model, $t(57) = 2.97$, $p < .01$. More recent reported symptom onset was associated with elevated perceptions of symptom severity in children relative to caregivers.

When entering the overall index of caregiver psychological symptoms (SCL-90-R Grand Total) on the subsequent step of the model predicting child–caregiver discrepancies in reports of symptoms severity, the change in variance accounted for was found to be significant, $R^2$ change = .06, $F(1, 53) = 4.05$, $p = .05$. Greater caregiver endorsement of personal psychological symptoms was uniquely associated with elevated perceptions of symptom severity in their children relative to child reports. Caregiver psychological symptoms also accounted for significant incremental proportions of variance in caregiver–physician symptom severity report discrepancies, $R^2$ change = .11, $F(1, 53) = 7.13$, $p = .01$, with caregivers endorsing more personal psychological symptoms having elevated perceptions of their children’s symptom severity relative to the physician. No model was found to be significantly predictive of child–caregiver and caregiver–physician report discrepancies on the other disease severity variables (i.e., perceptions of pain or functional limitation).

Results for the regression of informant discrepancies onto objective indicators of disease severity (after controlling for child age, gender, and reported age at symptom onset) indicated that the objective disease severity indicators together accounted for significant variance in caregiver-physician differences in perceived symptom severity beyond the control variables. The incremental change in variance accounted for was statistically significant, $R^2$ change = .15, $F(3, 54) = 3.52$, $p = .02$. Similarly, the objective indicators accounted for an additional 12.7% of the variance in child–physician differences in perceived symptom severity beyond the control variables; the incremental change in variance accounted for approached statistical significance, $R^2$ change = .13, $F(3, 54) = 2.66$, $p = .06$, and disease subtype was a significant unique predictor in the full model, $t(41) = 2.25$, $p = .03$. The direction of results indicated that the attending physician was more likely to perceive symptom severity as lower than the caregiver and child if the objective indicators of severity suggested less severe disease. No model was found to be predictive of child–physician and caregiver–physician report discrepancies on perceptions of pain or functional limitation.

**Discussion**

We found support for informant discrepancy in perceptions of disease-related symptom severity, with children on average reporting less severe disease than their caregivers and physicians. Discrepancies between caregivers and the other informants in part were found to be
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accounted for by the psychological adjustment of the caregiver, with poorer adjustment associated with perceiving higher symptom severity in their children relative to other informants. Discrepancies between physicians and the other informants (children and caregivers) in part were found to be accounted for by objective medical indices, with lower disease severity suggested by the medical indices being associated with lower physician symptom severity ratings relative to the other informants.

Our findings of significant child–caregiver discrepancies in perceptions of symptom severity are consistent with previous literature suggesting that caregivers tend to report more problematic psychological and behavioral symptoms for their children than the children report for themselves (Canning, 1994; Handwerk et al., 1999; Klein, 1991), and that other informants tend to maximize the seriousness of chronic disease in children relative to the child’s own report (Law, 2002). This finding further supports the need for multisource data in pediatric populations and may have implications for adherence to medical regimens (e.g., children may be less adherent if they perceive their disease to be less severe than do others).

The finding that caregiver endorsement of psychological symptoms was a significant predictor of child-caregiver and caregiver–physician discrepancies in perceptions of disease severity is in concordance with previous literature on the influence of caregiver psychopathology in exaggerating caregiver reports of child symptoms (Briggs-Gowan et al., 1996; Richters, 1992). Thus, taken together with previous research in this area, caregiver psychological symptoms appear to have a significant impact on the reporting of both psychological symptoms and the severity of disease-related symptoms in children. These data support the need for caregiver adjustment to be routinely measured in studies evaluating pediatric chronic illness, particularly if caregiver reports are used as a primary outcome measure. Further, these data appear to support the need for caregivers to be included in clinical interventions.

Of further interest to us was that physicians reported higher or lower disease severity relative to other informants based on the severity suggested by objective medical indices. Although no previous studies could be located that have explicitly addressed this issue in children, this finding is consistent with studies on adult chronic illness populations which suggest that informant discrepancy may in part be explained by physicians basing their perceptions largely on objective medical data (e.g., Abbott et al., 1995). Further, our findings suggest that the inclusion of physician disease severity ratings may provide an objective metric against which to judge whether children and caregivers are under- or over-reporting chronic disease severity.

The findings of this investigation must be interpreted within the limitations of the study design. The study is correlational in nature, and as such causality underlying relationships among variables cannot be determined. Indeed, some relationships may be bidirectional (e.g., between disease severity and caregiver adjustment). The use of difference scores as an outcome variable, though still common and adequate for scores on an identical metric, has been criticized by some (e.g., Holmbeck et al., 2002), and more advanced statistical procedures with larger samples may improve confidence in findings. Further, our findings are based on SCD as a chronic disease exemplar and primarily mothers as caregiver informants; the findings may, therefore, not generalize to other chronic disease populations or other caregiver informants (e.g., fathers). Finally, limited sample size was an issue for statistical power in the current study, and some of the intercorrelations among informant measures would have reached statistical significance in studies having a sample size of at least 100 participants (based on post hoc power analysis and an r-value of .25). Findings thus remain tentative until the study is replicated using other and larger chronic illness samples.

In conclusion, researchers investigating the relationship between severity of chronic disease and adjustment in children may be measuring different constructs depending on the informant. Future studies in this area should incorporate multiple informants and consider using statistical procedures for aggregating multisource data (see Holmbeck et al., 2002).

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