Brief Report: Predictors of Parenting Stress Among Parents of Children With Biochemical Genetic Disorders

Susan E. Waisbren,1 PhD, Michelle Rones,1 PhD, Catherine Y. Read,2 PhD, RN, Deborah Marsden,1 MD, and Harvey L. Levy,1 MD
1Children’s Hospital and Harvard Medical School, and 2Boston College School of Nursing

Objective To examine predictors of parenting stress in parents whose children were diagnosed with a biochemical genetic disorder clinically or through newborn screening. Methods Parents of 263 children with biochemical genetic disorders (139 identified by newborn screening, 124 identified clinically) completed interviews focused on child health, medical service use, satisfaction with services, parenting stress, and family functioning. Results Multiple regression analyses suggested that child adaptive functioning, parental satisfaction with support, and difficulties parents experienced meeting their child’s health care needs were associated with scores on the Parenting Stress Index ($R^2 = .51$). Conclusions Initiatives to improve child adaptive functioning and parental support as well as practical assistance to help parents meet their child’s health needs may reduce parental stress and family disruption in this population.

Key words biochemical genetic disorders; metabolic disorders; parenting stress; newborn screening.

Biochemical genetic disorders are characterized by disruption of normal metabolic functions affecting fatty acid oxidation, amino acids, organic acids, and the urea cycle. While individually these disorders are rare, as a group they account for substantial levels of pediatric morbidity and mortality (Ellaway, Wilcken, & Christodoulou, 2002; Yang, Khouyr, & Mannino, 1997).

Children with biochemical genetic disorders vary greatly in terms of health, treatment, and neurodevelopmental outcome. This variability is related to a combination of factors, including the nature of the enzyme deficiency and the timing and continuity of treatment (generally the earlier the better) (Filiano, Bellimer, & Kunz, 2002; Goodwin, Msall, Vohr, Rubin, & Padbury, 2002). Treatment of the metabolic disorder may consist of dietary restrictions/supplementation, increased frequency of feeding, and the monitoring of blood levels (Filiano et al., 2002).

Despite treatment, a significant proportion of children with biochemical genetic disorders have developmental disabilities (Calvo et al., 2000; “Mental Retardation Following Diagnosis,” 1999) and require heightened levels of parental support, supervision, and care. Given the genetic origin of these biochemical disorders, parents must also face difficult decisions regarding future pregnancies (Collins, Halliday, Kahler, & Williamson, 2001; Read, 2002).

Given the range of demands that parents of children with biochemical genetic disorders face, they are at high risk for parenting stress and its negative associated outcomes, including maternal anxiety and depression (Naerde, Tambs, Mathiesen, Dalgard, & Samuelsen, 2000), negative parent–child interactions (Morgan, Robinson, & Aldridge, 2002), insecure infant attachment (Atkinson et al., 2000), physical abuse (Rodriguez & Green, 1997), and child emotional and behavioral problems (Goldberg et al., 1997).

This report examines predictors of parenting stress in a large cohort of parents of children with biochemical genetic disorders. A previous report on the same cohort...
examined parental reproductive decisions (Read, 2002). Additionally, a small subset of this cohort was described in a brief report (Waisbren et al., 2002) which compared hospitalization rates, incidence of mental retardation, and parental stress among children with the same diagnosis who were identified by newborn screening \((n = 28)\) or clinical symptoms \((n = 17)\). Results suggested that while both groups experienced similar rates of hospitalization, children in the clinically diagnosed group had higher rates of mental retardation and their parents experienced higher levels of stress than the children diagnosed through newborn screening. The present study builds on these results through the examination of predictors of parenting stress, application of a theoretical model, and increase in sample size.

The Interaction Model of Client Health Behavior (Figure 1) (Cox, 1982) guided variable selection and data analyses. The model includes parent, child, medical system, and health outcome elements. It incorporates individual parent differences, focuses on the interaction between the parent and the health care system, and includes psychosocial as well as medical outcomes.

Based on the model, we hypothesized that parental stress would be predicted by child adaptive functioning, parental resources, and parent–provider relations.

**Method**

**Participants**

Parents of 263 children with biochemical genetic disorders (139 identified by newborn screening, 124 clinically) participated in the study. Families were recruited through the New England Consortium of Metabolic Programs (Albers et al., 2001), and 83% of those contacted agreed to participate.

Sample families were predominantly white (91%, \(n = 239\), with 6% Hispanic, \(n = 15\), and 2% African American, \(n = 5\)) and middle class. The majority of sample parents were married (75%, \(n = 197\)), with a median family size of two children. The child sample comprised roughly equal numbers of girls (142, 54%) and boys (120, 46%). On average, sample children were 8.5 years of age at the time of the study (range, 6 months to 18 years). Thirty-eight biochemical genetic disorders were

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**Figure 1.** Interaction model of client health behavior applied to the study of parenting stress in parents of children with biochemical genetic disorders.
represented, including phenylketonuria \((n = 112)\), galactosemia \((n = 26)\), arginosuccinic acidemia \((n = 4)\), glutaric acidemia types I and II \((n = 6)\), medium-chain acyl-CoA dehydrogenase deficiency \((n = 9)\), and maple syrup urine disease \((n = 6)\). Children identified by newborn screening were diagnosed by age 2 months compared with a median of 9.5 months \((\text{range, 0–11 years})\) in the clinically ascertained group.

**Procedure**

The New England Consortium of Metabolic Programs collaborated in developing a survey instrument, recruiting subjects, and implementing the study \((\text{Albers et al., 2001})\). Parents of children with metabolic disorders were recruited through letters and contacted by phone to complete an interview. One parent per family completed the interview. Sample parents were predominantly mothers \((89\%, n = 234)\); fathers, \(10\%, n = 26)\); and grandparent guardians, \(1\%, n = 2)\). Informed consent was obtained over the phone or in writing, as approved by the institutional review boards at each of the metabolic centers.

**Measures**

**Interview**

A structured interview was administered to obtain information on background characteristics, socioeconomic status \((\text{Hollingshead & Redlich, 1954})\), and diagnosis of the child. Several questions addressed social support \((\text{Pierce, Sarason, & Sarason, 1991})\). The number of people in the parents’ social support network was determined by having the parents name all the people they could count on when they needed help. Parental satisfaction with social support was assessed through parental response to the question “How satisfied are you with the overall support you receive?” Parents were asked to rate their responses on a 5-point Likert scale, ranging from 1 = not at all satisfied to 5 = very well satisfied. Difficulty meeting the child’s extra needs (related to his or her metabolic disorder) was assessed via parental response to a 5-point Likert scale ranging from 1 = not difficult to 5 = extremely difficult.

**Parenting Stress Index–Short Form (PSI-SF)**

The PSI-SF \((\text{Abidin, 1995})\), a 36-item index designed to assess stress in the parent–child system, was administered to parents. The PSI-SF has good internal consistency \((\text{coefficient alpha} = .90)\); for the present study sample, \(80)\), relates highly to the full PSI \((r = .92)\), and has been used extensively in research and clinical settings. In the present sample, 48 children were older than 12 years of age \((\text{the PSI was validated on children ages 1 month to 12 years})\). The PSI scores for the parents of children between ages 13 and 18 years were not significantly different from those of parents of children ages 12 and under.

**Vineland Adaptive Behavior Scale (VABS)**

The VABS \((\text{Sparrow, Balla, & Cicchetti, 1984})\) was administered to parents to measure child adaptation in various domains \((communication, daily living, social, and motor skills)\). The VABS composite score was used to measure the child’s developmental level.

**Results**

Summary data on the study variables for the entire sample as well as the newborn-screened and clinically identified groups are presented in Table I. Data for all 263 children \((139 \text{ newborn screened and 124 clinically identified})\) were available for all variables described.

**Parental Stress**

The mean score on the PSI-SF was 76 \((\text{range, 36–155})\), comparable to the normative mean. The distribution of scores on the PSI-SF was generally normal, but 32\% \((n = 84)\) of parents reported clinically significant levels of parenting stress \((above the 84th percentile)\).

<table>
<thead>
<tr>
<th>Variable</th>
<th>(M)</th>
<th>SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td>PSI-SF</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total sample</td>
<td>75.6</td>
<td>25.4</td>
<td>36–155</td>
</tr>
<tr>
<td>Newborn screened</td>
<td>64.9</td>
<td>18.4</td>
<td>36–117</td>
</tr>
<tr>
<td>Clinically identified</td>
<td>87.6</td>
<td>26.8</td>
<td>36–155</td>
</tr>
<tr>
<td>VABS composite score</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total sample</td>
<td>83.5</td>
<td>28.2</td>
<td>20–146</td>
</tr>
<tr>
<td>Newborn screened</td>
<td>97.5</td>
<td>17.3</td>
<td>43–146</td>
</tr>
<tr>
<td>Clinically identified</td>
<td>67.8</td>
<td>29.9</td>
<td>20–122</td>
</tr>
<tr>
<td>Difficulties meeting child’s needs</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Total sample</td>
<td>1.7</td>
<td>1.7</td>
<td>0–5</td>
</tr>
<tr>
<td>Newborn screened</td>
<td>0.9</td>
<td>1.3</td>
<td>0–5</td>
</tr>
<tr>
<td>Clinically identified</td>
<td>2.5</td>
<td>1.7</td>
<td>0–5</td>
</tr>
<tr>
<td>Satisfaction with social support</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
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<td>4.1</td>
<td>1.2</td>
<td>1–5</td>
</tr>
<tr>
<td>Newborn screened</td>
<td>4.4</td>
<td>1.0</td>
<td>1–5</td>
</tr>
<tr>
<td>Clinically identified</td>
<td>3.8</td>
<td>1.3</td>
<td>1–5</td>
</tr>
<tr>
<td>Number of people in support network</td>
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<td></td>
<td></td>
</tr>
<tr>
<td>Total sample</td>
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<td>4.2</td>
<td>0–21</td>
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<tr>
<td>Newborn screened</td>
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<td>4.1</td>
<td>0–21</td>
</tr>
<tr>
<td>Clinically identified</td>
<td>4.6</td>
<td>4.2</td>
<td>0–20</td>
</tr>
</tbody>
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PSI-SF = Parenting Stress Index–Short Form; VABS = Vineland Adaptive Behavior Scale.
Factors Associated With Parental Stress

Child Characteristics. Child developmental level correlated with parenting stress ($r = .63$, $p < .0001$) and was related to the timing of diagnosis and treatment ($r = -.30$, $p < .001$). Children who were newborn screened achieved significantly higher scores on the VABS composite score ($t = 9.98$, $p < .001$) compared with those identified clinically.

Difficulties Meeting Child’s Needs. Difficulties meeting the child’s health care needs also correlated with parental stress ($r = .63$, $p < .0001$). There were significant differences between the clinically identified and the newborn-screened groups. Among parents of clinically identified children, 30% ($n = 79$) reported significant difficulties meeting their child’s health care needs (defined as a score > 3 on the 5-point scale) compared with only 5% ($n = 13$) of parents of children in the newborn-screened group ($\chi^2 = 28.94$, $p < .001$). A significantly greater percentage of clinically identified children required hospitalizations ($\chi^2 = 75.75$, $p < .001$), assistive devices (wheelchair, gastrostomy tube) ($\chi^2 = 89.03$, $p < .001$), and specialized services ($\chi^2 = 21.96$, $p < .01$).

Social Support. The number of people in the parents’ social support network and parental satisfaction with social support were related to parenting stress ($r = .37$, $r = .38$, respectively, $p < .0001$). Parents of clinically identified children listed fewer people in their social support network ($t = 2.35$, $p < .02$) and reported less satisfaction with the support they received ($t = 4.18$, $p < .001$).

Predictors of Parental Stress

Variables that were significantly related to parental stress were entered into the regression equation. The total model yielded an $R^2$ of .51, $F(3, 259) = 89.88$, $p < .001$. The VABS composite score was entered into the stepwise regression equation first and had an $R^2$ of .39. Next, the extent to which parents experienced difficulties meeting their child’s health care needs was entered, which increased the $R^2$ by .11. Then, parental satisfaction with the social support network was entered, which increased the $R^2$ by .01. Whether or not the child was diagnosed through newborn screening was also significantly associated with the score on the PSI-SF ($r = .45$, $p < .0001$), but because of its correlation with the VABS score ($r = .53$, $p < .0001$), it dropped out of the regression model.

Discussion

Child adaptive functioning, parental satisfaction with social support, and difficulty meeting the child’s health care needs predicted parenting stress. Together, these variables explained 50% of the variance in parenting stress among parents of children with biochemical genetic disorders. These results are consistent with prior research on the impact of child health problems and developmental disabilities on parenting stress (Halpern, Brand, & Malone, 2001; Pelchat et al., 1999; Trute & Hiebert-Murphy, 2002).

Child adaptive functioning and parental stress were associated with the timing of diagnosis and initiation of treatment. Compared with the clinically identified children, children diagnosed through newborn screening demonstrated significantly higher levels of adaptive functioning and fewer hospitalizations, and their parents reported significantly lower levels of parenting stress, fewer difficulties meeting their child’s health care needs, and higher levels of social support.

The results of the present study must be interpreted in the context of its limitations. The study was retrospective in nature, heterogeneous in its sample (both diagnostically and in terms of child age), and based only on parental reports. Additionally, because the study was cross-sectional, it is impossible to assess directionality.

Nonetheless, the study supports the notion that earlier diagnosis is associated with improved child functioning, which in turn is related to reductions in parental stress. Additionally, the study suggests that factors relating to both parents and children (e.g., social support networks, perceived difficulty meeting the child’s extra health care needs, and child adaptive functioning) are predictive of parenting stress. Therefore, initiatives targeted to each of these areas may result in reductions in stress for parents of children with biochemical genetic disorders.

Future research in this area should examine factors associated with parental stress for each biochemical genetic disorder (e.g., maple syrup urine disease) or class of disorders (e.g., fatty acid oxidation dysfunction) separately. Different factors may be more predictive of parental stress in particular disorders or classes of disorders because of differences in availability, effectiveness, and ease of treatments for particular disorders. Additionally, future research efforts should investigate the aspects of caring for a child with a biochemical genetic disorder that parents deem especially difficult and stressful (e.g., demands of frequent feeding, infant irritability, dietary compliance, medical appointments, activities of daily living).

Through a more complete understanding of the factors that make caring for a particular child more stressful for a particular parent, researchers can design intervention programs aimed at reducing parental stress...
and improving child functioning and quality of life. The next wave of research might also include thorough evaluations of various initiatives aimed at reducing parental stress in this population (e.g., physician education, parent education, support groups for parents of children with rare disorders).

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