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Mothers' and Fathers' Reports of Stress in Families of Infants With and Without Single-Suture Craniosynostosis

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Abstract

Objective—To compare relative levels of stress reported by mothers and fathers in families containing infants with and without single-suture craniosynostosis.

Design—Case-control study.

Participants—Mothers and fathers of 246 infants with recently diagnosed single-suture craniosynostosis and 253 frequency-matched control infants completed the Parenting Stress Index just prior to their infant's cranioplastic surgery. Family demographic information and mothers' ratings of the severity of their child's single-suture craniosynostosis were obtained.

Results—Average Parent Domain scores for parents of infants with single-suture craniosynostosis differed little from those reported by parents of control infants; however, Child Domain scores among parents of infants with single-suture craniosynostosis were higher on some subscales, primarily related to unexpected infant health and appearance issues. In both groups, fathers reported higher Child Domain stress than mothers, and mothers reported higher Parent Domain stress than fathers. Case mothers reported greater stress if they perceived their child's condition as more noticeable to others.

Conclusions—Prior to cases' cranioplastic surgery, parents of children with and without single-suture craniosynostosis reported similar levels of stress in relation to their parenting roles and the behavioral characteristics of their infants. Visibility of condition should be considered a risk for

increased stress for mothers of infants with single-suture craniosynostosis. Stress differences between mothers and fathers were far more discernible than those associated with the presence or absence of single-suture craniosynostosis.

Keywords

craniofacial condition; craniosynostosis; infancy; parenting stress

The birth of an infant is typically a joyous occasion but also represents a time of significant transition, when the expanding family is required to adjust to new roles and demands (Kazak and Marvin, 1984; Goldberg et al., 1990; Gottman and Gottman, 2007). The birth of any infant can be stressful for family members, but particularly infants with serious medical conditions (Goldberg et al., 1990; Cohen, 1999; Lightsey and Sweeney, 2008). Several factors can affect family adjustment when an infant has special medical needs, including uncertainty regarding the child's immediate condition and long-term prognosis, severity of the condition, frequency of medical appointments, time needed for therapies, and increased financial burden (McKinnery and Peterson, 1987; Cohen, 1999; Macias et al., 2007).

The diagnosis of a medical condition in an infant can therefore be considered a major life stressor, precipitating numerous minor and major crises that affect the family (Crnic et al., 1991; Halpern et al., 2001). Several models have been proposed to explain the link between these stressors and adaptation in families (see, for example, Patterson, 1998; Thomson et al., 1993a, 1993b; Wallander and Varni, 1998), including the idea that stressors are precipitating events that can disrupt family equilibrium, leading to a positive or negative change in family functioning (Cherry, 1989; Patterson, 2002; Folkman, 2008).

Measures of parent-related stress have been commonly used as markers of the degree to which a parent has been affected by the demands of the role. Many studies have examined parental stress in relation to children who have medical conditions (e.g., mental retardation, cardiac problems, cystic fibrosis, cerebral palsy, diabetes, spina bifida, very low birth weight), with most finding that parents of a child with a diagnosed medical condition report more stress and problems with adaptation than parents of children without such conditions (Kazak and Marvin, 1984; Britner et al., 2003; Nereo et al., 2003; Uzark and Jones, 2003; Hung et al., 2004; Vermaes et al., 2007; Wulffaert et al., 2009). There is also some evidence that higher parenting stress is associated with poorer child outcomes over the long term, including both internalizing and externalizing behavior problems (Anthony et al., 2005; Pope et al., 2005; Pai et al., 2006; Ashford et al., 2008). However, other studies have suggested that the stress associated with parenting a child with a medical condition can activate parental coping resources that promote resilience in the child and family (Case-Smith, 2004; Vermaes et al., 2007; Lightsey and Sweeney, 2008; Baker et al., 2009; Feragen et al., 2009).

Single-Suture Craniosynostosis

In the current study we examined parents' psychological responses to having an infant with single-suture craniosynostosis (SSC), a relatively common craniofacial condition in which a single skull suture is prematurely fused. Single-suture craniosynostosis results in an abnormal skull shape and has a possible, but uncertain, impact on neurodevelopmental functioning, presumably due to the possibility of increased cranial pressure or morphological changes in the brain (Speltz et al., 2004; Kapp-Simon et al., 2007). Cranioplastic surgery to release the fused suture is typically recommended with the dual goal of reshaping the skull to normalize appearance and minimizing neurodevelopmental impact. This procedure is generally performed in the first year of life; it has a mortality rate

approaching 1% and a morbidity rate of about 7% (Sloan et al., 1997). Due to the impending surgery and possible uncertainty about the child's long-term functioning, parents may experience relatively high levels of stress, particularly during the diagnostic process and prior to surgery.

In some previous studies, mothers of children with craniofacial disorders have reported elevated levels of parenting stress and more caregiving difficulties than mothers of healthy children (Field and Vega-Lahr, 1984; Wasserman and Allen, 1985; Barden et al., 1989; Speltz et al., 1990; Speltz et al., 2004; Paley et al., 2006). Research on craniofacial conditions has also shown that lower levels of parenting stress are associated with better child adjustment (Krueckeberg and Kapp-Simon, 1993; Krueckeberg et al., 1993; Pope et al., 2005) and better parental health status (Coulter et al., 1991). Baker et al. (2009) documented that positive, approach-oriented coping strategies were correlated with better parental adjustment for parents of children with cleft lip and palate. The types of coping strategies indexed by Baker et al. have been associated with lower levels of caregiving stress (Folkman, 2008; Park, 2010).

Whether parent functioning is viewed primarily in terms of resilience or risk for poor outcomes, parents' cognitive, affective, and behavioral responses to stressful events must all be considered (Patterson, 1983; Deater-Deckard, 1998; Crnic et al., 2005; Pope et al., 2005; Haskett and Willoughby, 2007; Kwon, 2007; Baker et al., 2009). Abidin's (1992, 1995) model of parenting stress is a good example of a multifaceted approach, focusing on parental perceptions of and attributions about the child's various characteristics, the quality of the parent-child and marital relationships, and parents' roles and responsibilities.

In this study we assessed the psychological impact of SSC on the family by measuring self-reported parenting stress levels in the time period between the infant's diagnosis and surgery. Parenting stress reports were also completed by parents of infants who did not have craniosynostosis or any other known medical condition. We hypothesized that parents of infants with SSC would report more parenting stress than parents of infants without a medical condition.

Both mothers' and fathers' perceptions of parenting stress were assessed. In the few studies of pediatric populations including the stress reports of both parents, fathers of children with medical conditions tended to report the same elevated levels of global stress reported by mothers (Goldberg et al., 1990; Roach et al., 1999; Macias et al., 2007), but the type of reported stressor differed by parent gender (e.g., stress related to child characteristic versus personal or familial issues; Beckman, 1991; Krauss, 1993; Esdaile and Greenwood, 2003; Butcher et al., 2008). Based on these findings, we hypothesized that mothers and fathers would report distinct patterns of adaptation related to differences in mothering and fathering roles.

Among families containing infants with SSC we also examined whether parenting stress was related to the affected suture (e.g., sagittal versus metopic synostosis) and to mothers' perceptions of their infants' craniofacial appearance. Single-suture craniosynostosis varies considerably in the extent to which the condition is visible or noticeable to others. However, objective ratings of a subgroup of the subjects in this study by individuals unfamiliar with the children demonstrated that the infants with SSC were rated as less attractive than a comparison group of same-aged children without SSC (Collett et al., 2009). Parents whose children look different may be subjected to questions about their child's appearance or may perceive that others look at their child in a less favorable manner, which can be upsetting (Rumsey and Harcourt, 2007). It was therefore hypothesized that increasing visibility of SSC would be associated with higher parenting stress.

To summarize, the aims of the study were as follows: (1) to examine differences in parenting stress levels between the case and control groups, (2) to examine differences in parenting stress levels of mothers and fathers regardless of group, and (3) among families of infants with SSC, to evaluate the relation between maternal perceptions of infants' craniofacial appearance and parenting stress levels.

Method

Participants

As part of a larger study examining neurobehavioral functioning of children with craniosynostosis, the parents of infants with craniosynostosis and parents of infants in the control group were asked to complete a measure of parenting stress. Participants in the larger study included 266 infants with SSC (cases) and 259 frequency-matched infants without craniosynostosis (controls) and their parents. All parents who completed the parenting stress measure (246 mothers and 210 fathers of cases and 253 mothers and 220 fathers of controls) were included in the current analyses. Participants were enrolled between 2001 and 2006, after obtaining informed consent approved by the institutional review boards (IRBs) of the institutions participating in this research, which was in full compliance with Health Insurance Portability and Accountability Act standards. This was a multi-institutional study, which included five participating sites. Each participating center obtained independent institutional approval of the study protocol and approval to share de-identified data across research sites.

Cases—Infants with SSC were referred to the project at the time of diagnosis by the treating surgeon or pediatrician. Infants were eligible to participate if they (1) had confirmed SSC (sagittal, metopic, unilateral coronal, or unilateral lambdoid synostosis), (2) were under 30 months of age, and (3) had not yet had corrective surgery. Infants with SSC were excluded if they (1) were born before 34 weeks' gestation, (2) had major medical or neurological conditions (e.g., cardiac defects, seizure disorders, cerebral palsy, significant health conditions requiring surgical correction), (3) had three or more extracranial minor malformations as defined by Leppig et al. (1987), or (4) had any other major malformations.

The 266 cases enrolled in the larger study represent 82% of all those referred and eligible. Forty-eight families (15%) declined to participate. The majority of nonparticipants (43%) did not respond to our invitation after referral by their treating physician. For those families eligible to participate who actively declined, the most commonly identified reason was lack of interest (33%). Other reasons provided included time and distance (8.2% each) and confidentiality concerns (2%). Among the 266 cases enrolled in the larger study, 246 mothers and 210 fathers completed the Parenting Stress Index (PSI) (Abidin, 1995) between the time of diagnosis and surgery. There were 194 cases who had PSI protocols available from both parents. For 18 cases, neither parent completed the PSI. Additionally, PSI protocols were missing from two mothers and 37 fathers.

Controls—Infants were eligible as controls if they had no known craniofacial condition and did not meet any of the exclusionary criteria for infants with SSC. Infants with isolated minor medical conditions such as colic, acid reflux disease, or allergies were eligible. Infants were recruited as controls who were generally similar to case infants, taking into consideration age, gender, family socioeconomic (SES) status within the same Hollingshead (1975) four-factor classification category, and ethnicity. A pool of 581 control group participants were recruited through pediatric practices, birthing centers, and announcements in newsletters and/or other publications of interest to parents of newborns.

The 259 control group participants who were enrolled in the larger study represented 44% of all those who were eligible. The largest percentage of nonenrolled controls were those who were on our waiting list of interested participants, but the control infant aged out of the project before a matching case was enrolled (76% of interested controls). Of eligible controls, 12% did not respond when contacted after a match was made. Other reasons for nonparticipation included time (4%), distance (3%), lack of interest (3%), and confidentiality concerns (2%). Among the 259 control group infants enrolled in the larger study, PSI data were available for 253 mothers and 220 fathers. The PSI data were missing from both parents for one control group infant, from mothers of two additional infants, and from fathers of 19 others.

The current analyses pertain to the 246 cases and 253 controls for whom either parent completed the PSI.

Measures

Parenting Stress Index—The PSI (Abidin, 1995) is a 101-item self-report questionnaire that assesses parenting stress and the presence and/or intensity of potential stressors in the family environment using 5-point Likert scales (*strongly agree* to *strongly disagree*). The PSI is widely used, has been included in approximately 800 published studies since 2001, and has been translated into 40 languages (Abidin, personal communication, 2010). It yields three global scores: Parent Domain, Child Domain, and Total Stress. The Parent Domain score includes seven subscales measuring parents' personal characteristics and social support as they relate to the demands and tasks of parenting: Depression, Attachment (to child), Restriction of Role, Competence, Isolation, Health, and Spousal Relationship. Similarly, the Child Domain score yields six scores related to parent appraisal of child characteristics or temperament, represented by the following subscales: Adaptability, Acceptability, Demandingness, Mood, Distractibility, and Reinforces Parent. Higher scores reflect higher levels of stress or greater presence and/or intensity of stressors in a given category; for example, *less* adaptive child behavior, *less* child reinforcement of parent, *greater* child demandingness, parent isolation, or parent depression are represented by higher scores on these subscales. Scores over the 85th percentile on the domains (148 on Parent Domain [range, 69 to 188]; 116 on Child Domain [range, 50 to 145]; and 258 on Total Stress [range, 131 to 320]) suggest a stress level that may adversely impact parenting or difficulties in behavioral and temperamental qualities of the child. There is also an additional Life Stress scale that assesses the presence of 19 potentially stressful life events in the past year (e.g., divorce, separation, death of family member or friend).

The PSI has been validated in several populations, including children with developmental issues, behavior problems, disabilities and illnesses, and high-social-risk families (Abidin, 1995). The instrument has been highly correlated with various measures that examine depression, behavior, and family impact. Cronbach alphas for sub-scales in the standardization sample ranged from .70 to .83 and were above .90 for the Parent, Child, and Total Domain scores. Test-retest reliability indicating the stability of scores across a 1- to 3-month interval was .63 for the Child Domain, .91 for the Parent Domain, and .96 for the Total Stress score (Abidin, 1995). We also calculated Cronbach alphas for the current sample. The domain scales (Parent, Child, and Total) had alphas of .91, .89, and .93, respectively. Subscale alphas ranged from .60 to .77, with one exception ($\alpha = .52$ for Distractibility/Hyperactivity, which may reflect the age of our infants). Equivalent alphas were found for cases and controls and mothers and fathers.

Maternal Perception of Condition Visibility—As part of a structured interview, mothers were asked to rate the degree of visibility of their infants' differences in craniofacial

appearance. Maternal visibility ratings ranged from 1 (*mother can not detect any difference*) to 5 (*the difference is noticeable to everyone*). To increase the number of subjects in each category, the scale was collapsed to a 3-point scale: 1 (*not or barely noticeable*), 2 (*noticeable to some people*), or 3 (*noticeable to everyone or almost everyone*) for analysis.

Procedure

The authors obtained informed consent following the IRB-approved protocols of each participating institution. Participants and their families were seen for evaluation between diagnosis and the child's surgery, on average 46.5 days prior to surgery. Mothers were interviewed to obtain medical history and family constellation data, and they completed the visibility ratings. The PSI was completed independently by each case and control parent, generally at home, and returned by mail.

Data Analyses

Scores for cases and controls were summarized separately for mothers and fathers by calculating means and standard deviations after adjusting for infants' age at time of evaluation, race/ethnicity (white versus other), gender, study site, and SES.

The study hypotheses were evaluated by using a generalized estimating equations (GEE) approach applied to linear regression models (Hardin and Hilbe, 2003). All regression models were adjusted for SES (Hollingshead, 1975; continuous variable), infant age when evaluated (in months), race/ethnicity (dichotomized as white/other due to the small representation of nonwhite subjects), infant gender, and study site. These variables were originally theoretically chosen as matching variables. They were entered as covariates to account for the fact that participants were ultimately frequency matched on these demographic characteristics. In addition, adjusting scores allowed us to alleviate any potential confounding of group differences by these demographic characteristics. For each estimate derived from regression models, 95% confidence intervals (CI) and *p* values were calculated. Because this study had few specific hypotheses about how stress in various subdomains would differ among case and control mothers and fathers, most analyses were considered exploratory and did not adjust *p* values for multiple comparisons. All CIs and *p* values are reported in order to allow full interpretation of the findings in the context of the study's variables and design.

Differences Between Cases and Controls and Between Mothers and Fathers—

To evaluate whether mothers and fathers of infants with SSC had higher levels of stress than mothers and fathers of controls, we fit a linear regression model by using GEE with repeated measures (i.e., each infant had scores for both mother and father). First, case-control group differences for mothers and fathers were estimated separately (i.e., comparing case mothers with control group mothers, case fathers with control group fathers). Next, we examined whether the magnitude of mother-father differences differed for cases and controls by including terms for case status, parent's gender, and the interaction between case status and parent gender in the regression model.

To determine whether there were case-control group differences in the source of stress reported by mothers and fathers, we repeated the GEE with case status, type of stress (parent- or child-related), and the interaction between the two. The interaction term indicated whether the domain in which greater stress was identified differed for cases and controls.

Associations Between Parenting Stress and SSC Diagnosis Group and Mothers' Perceptions of Appearance—

In order to evaluate whether SSC diagnosis or

maternal ratings of infants' craniofacial appearance were predictive of stress levels, linear regression analyses were performed for cases only. In these models, independent variables included the matching factors (SES, age at evaluation, child gender, race/ethnicity, study site) as well as diagnostic group (sagittal, metopic, or unicoronal), and maternal ratings of SSC visibility.

Results

Demographic Characteristics

The majority of cases (64%) and controls (64%) were boys, due to the higher frequency of boys in the most commonly occurring craniosynostosis diagnoses (e.g., sagittal and metopic; Table 1). The average age was the same for both cases and controls (7.3 months). Average SES was upper middle class for both groups; however, infants with SSC were less often from families of higher SES (62% in Hollingshead levels I and II as compared with 80% of controls). This difference may be due to a recruitment bias: Control families with greater resources may have been more willing to participate. The majority of infants lived with their biological mothers and fathers, with 6.5% of cases and 3.2% of controls living only with their mothers. Both case and control families typically had one (35% and 46%, respectively) or two children (38% and 35%, respectively).

Differences Between Cases and Controls

Mothers—On average, case mothers had higher Child Domain total scores than control mothers (difference = 3.2, $p = .03$) and higher scores on most Child Domain subscales, although the differences were generally quite small (Table 2). The largest Child Domain subscale differences between case and control mothers were observed on the Demanding subscale (2 points higher) and Acceptability subscale (1.5 points higher, both $p < .001$).

Control mothers on average had higher Parent Domain total scores than case mothers and greater stress on all scales except one (Competence), although the differences were often minimal (Table 2). Parent Domain differences between case mothers and control mothers were most pronounced for the Role Restriction and Spousal Support subscales (both differences ~1 point, $p < .03$ for both comparisons). Control mothers also reported slightly more Life Event–related stress than case mothers (difference = 0.5, $p = .52$).

Fathers—Similar to the mother findings, case fathers had higher Child Domain total scores than control fathers (difference = 3.0, $p = .06$) and higher levels of stress on most Child Domain subscales, although the differences were small (Table 3). The largest mean differences between case and control fathers were found on the Child Demanding subscale (1.8 points higher, $p < .001$) and Acceptability subscale (1 point higher, $p < .01$).

Control fathers scored 1 point higher than case fathers on the Parent Domain total score ($p = .62$). Means for case and control group fathers on the Parent Domain subscales were nearly equal. Case fathers reported slightly more Life Event stress than control fathers (mean difference = 0.6, $p = .41$).

Differences Between Mothers and Fathers

Differences between mothers and fathers separately for cases and controls are shown in Tables 4 and 5. For the Child Domain, both case and control fathers generated higher total scores than case and control mothers (difference ~4 points for each, p values $< .001$). Fathers from both groups had slightly higher scores than mothers on every Child Domain subscale. For case fathers the largest differences were found on Adaptability and Reinforces Parent (differences ~1.0 to 1.6, $p < .001$) and Acceptability and Mood (differences ~0.5, p

< .04). For control fathers the largest differences were found for Reinforces Parent, Acceptability, and Adaptability (differences ~ 1.0 , $p < .01$), and Distractibility (difference ~ 0.6 , $p = .05$).

For the Parent Domain, case mothers' total stress scores averaged almost 3 points higher than case fathers ($p = .06$), and among control group participants, mothers scored 5 points higher than fathers ($p < .001$). On the Parent Domain subscales, both case and control mothers reported greater stress than fathers on five of the seven scales. Case and control mothers reported higher stress on the scales of Health, Role Restriction, Depression, and Spousal Support (case mothers' Role Restriction and Health was 0.8 higher, $p < .02$; Depression and Spousal Support ~ 1.5 higher, p values $< .001$; control mother values similar in magnitude except for Spousal Support, which was 2.5 points higher than the score for fathers, $p < .001$; see Table 5). Case and control fathers reported higher stress on the Attachment scale (differences ~ 1.5 , p values $< .001$).

On the Life Events scale, there were only slight mother-father differences among cases in the frequency of reported stressful events (difference = -0.5 , $p = .34$). Control group mothers reported higher levels of stress than fathers in the control group (1.6 points higher, $p = .01$).

Case-Control Group Differences in Mother-Father Differences—In addition to estimating mother-father differences separately for case and control parents, statistical tests of gender differences by case-control status were also performed. The patterns of mother-father differences were very similar among case and control parents, and the magnitude of these differences (by gender) did not differ by case-control status.

Associations Between Parent Stress and SSC Diagnostic Group and Perceived Visibility Child Domain—After adjusting for potential confounding factors of SES, infant age at assessment, gender, race/ethnicity, and study site, there were few differences in child-related stress for case mothers or fathers on the basis of diagnosis. With regard to visibility, mothers' reports of child-related stress were on average more pronounced among those who viewed SSC as highly noticeable compared with those for whom the condition was not generally noticeable (difference = 9.3; 95% CI = 3.1 to 15.4, $p < .001$).

Parent Domain—In a similar model where potential confounding factors were adjusted, on average parents of infants with sagittal and unicoronal synostosis reported similar levels of parent-related stress ($p > .95$ for mothers and fathers); whereas, parents of infants with metopic synostosis scored nearly 4 (mothers) or 7 (fathers) points less stress than parents in the other two groups (95% CI = -11.4 to 3.5 ; $p = .30$ for mothers; 95% CI = -14.1 to 0.8 , $p = .08$ for fathers). Parent Domain stress differed less in relation to SSC visibility, with stress reported by mothers who believed that the child's condition was noticeable to most everyone scoring 2.6 points greater than those reporting the condition as unnoticeable ($p = .52$). When the child's condition was viewed as highly noticeable by mothers, fathers reported less stress (1.6 points lower, $p = .70$) than those for whom the condition was viewed as "not or barely noticeable."

Discussion

In the current study the authors sought to determine whether the experience of having an infant with SSC generated parenting stress in excess of that associated with the parenting of a same-aged infant without medical illness or physical malformation. Parents' reports of psychological functioning and associated perceptions and attributions were gathered at a

point in time when one might expect particularly high levels of stress among parents of infants with SSC: soon after the infant's initial craniofacial diagnosis and prior to the infant undergoing a potentially life-threatening surgery. A second goal of the study was to examine differences between mothers and fathers, expanding upon the relatively few studies of medically compromised pediatric populations that included fathers as well as mothers. The third goal was to look at within-group differences by visibility of the synostosis from the mother's perspective.

Parents of infants with SSC reported slightly higher levels of child-related stress than did parents of healthy infants. However, for both mothers and fathers, these differences were confined almost entirely to two of the six Child Domain subscales (Demandingness and Acceptability). *Post hoc* inspection of the items composing these scales indicated that cases and controls had similar mean ratings across items with two exceptions: item 45 on the Demandingness scale ("My child has had more health problems than I expected") and item 21 on the Acceptability scale ("My child looks a little different than I expected and it bothers me at times"). Indeed, much of the overall difference between the groups on these scales is attributable to these two items, as indicated by the fact these differences decreased by ~1 point when the items were removed from analysis. These findings suggest that the primary reason for the higher child-related stress reported by parents of infants with SSC in this study was the violation of a common assumption: that one's infant will be physically normal without need in the first months of life for a major medical intervention such as cranioplasty.

The lack of parent group differences on Child Domain subscales may also relate to the relatively normal status of infants with SSC at this early point in the first year of life. Unlike infants with many other chronic medical conditions or physical disabilities, infants with SSC are generally healthy, and they do not have developmental delays so severe that they are obvious to most parents (see Speltz et al., 2007, for information on developmental functioning at the initial assessment). Because the Child Domain scales relate primarily to parent perceptions of infant temperament and behavior (Torowicz et al., 2010), comparable scores between control and case parents indicate that these infants are not, at least prior to surgery, unusually irritable or challenging to care for. Parents may feel stressed by the idea of surgery and they may have concerns about their infant's long-term prognosis, but they appear not to experience extraordinary stress attributable to infant behavior.

Even though on most dimensions of stress that relate to parent factors, mothers of infants with SSC reported less stress than mothers of healthy infants, these differences were most pronounced on the dimensions of Role Restriction and Spousal Support. These scales contain items describing the potentially negative impact of new parenthood on personal freedom (e.g., "Since having this child, I have been unable to do new and different things") and on one's relationship with a spouse or other partner (e.g., "Since having a child, my spouse and I don't do as many things together"). The fact that mothers of infants with SSC showed less frequent endorsement of these items suggests that having an infant with a serious medical condition—and associated events such as obtaining a medical diagnosis and preparing for surgery—may suppress or postpone the perceived limiting effects of parenthood on self and partner (e.g., Gottman and Gottman, 2007; Claxton and Perry-Jenkins, 2008). That is, mothers' attention may be directed solely toward the medically vulnerable infant, with less concern, at least initially, about the effects of parenting on adult roles and relationships. In addition, the fact that many fathers often accompany mothers to their child's doctor's appointments may provide more opportunities for parents to spend time together. It is also possible that social support from individuals outside the parent dyad may be higher during the period immediately preceding surgery. Studies have demonstrated that in families of children with a medical condition, higher levels of social support are

positively correlated with parent adjustment and more effective family coping (Visconti et al., 2002; Baker et al., 2009).

The findings in this study of little difference between the parents of children with and without SSC may also speak to the resilience of parents of children with SSC. Although these parents acknowledged their disappointment with their child's medical condition and appearance, they viewed their child and their family's functioning in a positive light overall. Their relatively low levels of reported stress may also reflect mutual parent support during a time of acute distress. The majority of these parents also received their child's care within the context of a multidisciplinary craniofacial team, and they may have benefited from support, education, or other resources from craniofacial team members.

Differences Between Mothers and Fathers

The observed differences between mothers and fathers in this study, regardless of infant status, were more striking and consistent than those between cases and controls. For both cases and controls, fathers reported higher child-related stress than did mothers, and mothers reported higher parent-related stress than did fathers. There was one exception to the latter finding: fathers in both groups (cases and controls) had higher scores than mothers on the Parent Domain Attachment subscale, which is consistent with previous research (Shonkoff et al., 1992). This subscale contains items pertaining to not only the parent's perception of closeness to the infant, but also comfort with holding and taking care of children, an area in which men have traditionally been viewed as less experienced. This explanation may also account for fathers' report of higher stress in relation to their child's temperament and other behavioral characteristics.

On other Parent Domain subscales, mothers of both cases and controls tended to report greater stress than their male counterparts on the Health, Role Restriction, Depression, and Spouse subscales. The items on these scales generally reflect the degree to which parenthood is viewed as having negative impact on one's physical and mental health as well as on relationships with family members, including spouses or partners. The greater stress among mothers than fathers, regardless of whether one is parenting an infant with a serious medical condition such as SSC, could be due to mothers' typically increased responsibility for child care (Beckman, 1991; Sloper et al., 1991; Moses et al., 1992; Roach et al., 1999). It is possible that, in particular, the Depression and Health subscale differences between mothers and fathers in this study are related to the depressive symptomatology, sleep disturbance, and other psychological and emotional difficulties experienced by many new mothers in the first few weeks or months of the postpartum period (Wolfson et al., 2003; Segre et al., 2007). In a Danish study, the risk of postpartum mental disorders among mothers was increased for several months after childbirth, but among fathers there was no excess of psychiatric disturbance during the same time period (Munk-Olsen et al., 2006).

It was expected that increased visibility of the condition from the mother's perspective would be associated with a parent's reports of higher parenting stress. Mothers of infants with SSC reported higher stress when they perceived their child's condition as more noticeable to others. Visibility of the condition is not directly related to diagnostic category; rather, each diagnostic category can have infants with greater or lesser severity and related visibility. A child with an atypically shaped head can draw negative attention to families, and this scrutiny may increase maternal anxiety and perceived stress. In addition, more severe head malformation may raise maternal concerns about possible adverse effects on the infant's brain development.

This study, while contributing noteworthy research to the craniofacial literature, has several limitations. Though widely used and reported in the craniofacial literature as well as

literature on children with medical conditions and physical disabilities, the PSI is a general measure of parenting stress in typical families, with no specification of particular medical or other conditions or disorders that affect infants. The PSI may not have captured the unique challenges of parenting a child with a craniofacial condition or a child in need of impending major surgery. Although we know of no such measure of craniofacial-specific parenting stress, this may be an important goal for future research. In addition, we did not directly assess coping strategies or social support, and these processes may have mediated the stress response of the parents in this research. Finally, the fact that the data were based only on self-reported experiences and processes raises the possibility of bias attributable to unknown factors.

Summary

Despite the plausibility that craniosynostosis-related diagnosis, surgical treatment, and infant appearance represent extraordinary stressors in the lives of new parents, the parents of infants with SSC in this study reported levels of stress very similar to those reported by parents of same-aged infants without medical illnesses, surgeries, or physical malformations. One exception is slightly higher child-related stress among parents of infants with SSC, related primarily to unexpected infant health and appearance issues. These findings may point to the resiliency of the parents of infants with SSC or, possibly, to a suppressed or delayed stress response related to the effects of caring for an infant requiring major cranial surgery.

Parents with and without infants having SCC showed a consistent pattern in which fathers reported higher child-related stress and mothers reported higher parent-related stress. Differences between mothers and fathers were more discernible than those associated with an infant diagnosis of SSC.

Although these findings suggest that most parents of infants with SSC cope effectively with their child's condition, other studies have found elevated levels of parenting stress in some families of children with various craniofacial conditions (Speltz et al., 1990; Pope and Ward, 1997) and such elevations, especially when they continue across time, have been associated with poorer child adjustment and social competence (Krueckeberg and Kapp-Simon, 1993, 1997; Pope et al., 2005). The planned longitudinal follow-up of these subjects will permit assessment of changes in stress over time, as well as the relationships between parenting stress and child behavioral development.

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TABLE 1

Demographic Characteristics of Single-Suture Craniosynostosis Cases and Controls at Time 1 Visit (Presurgery for Cases)

Characteristics	Diagnosis			Cases		Controls		
	Sagittal	Metopic	Unilateral	Lambdoid	n	%	n	%
Total number	114	58	62	12	246	49	253	51
Female	26	16	41	6	89	36	91	36
Male	88	42	21	6	157	64	163	64
Ethnicity								
Other	27	17	16	2	62	25	74	29
White	87	41	46	10	184	75	179	71
SES*								
I (high)	26	11	13	5	55	22	69	27
II	44	21	22	4	91	37	124	49
III	19	11	17	2	49	20	34	14
IV	20	10	5	0	35	14	15	6
V (low)	5	5	5	1	16	7	9	4
Site								
Seattle, WA	43	22	20	5	90	37	93	37
Chicago, IL	27	24	34	4	89	36	96	38
St. Louis, MO	21	2	6	2	31	12	29	11
Atlanta, GA	23	10	2	1	36	15	35	14

* SES = socioeconomic status, which is based on Hollingshead (1975).

TABLE 2

Mothers' Mean Parenting Index (PSI) Scores for Cases (n = 246^{*}) and Controls (n = 253^{*})

	Cases		Controls		Case-Control Difference [†]	
	Mean	SD	Mean	SD	Difference	95% CI
Child subdomains						
Distractibility	24.7	4.1	24.9	4.0	-0.2	-0.9 to 0.5
Adaptability	23.6	5.5	23.6	5.0	0.0	-0.9 to 1.0
Reinforces Parent	8.1	2.5	8.0	2.0	0.1	-0.3 to 0.5
Demanding	17.5	4.2	15.5	4.1	2.0	1.2 to 2.7
Mood	8.7	3.1	8.8	2.6	-0.1	-0.6 to 0.4
Acceptability	11.8	3.7	10.3	2.8	1.5	0.9 to 2.1
Parent subdomains						
Competence	25.2	5.5	25.1	5.1	0.2	-0.8 to 1.1
Isolation	12.3	4.0	12.6	3.5	-0.4	-1.1 to 0.3
Attachment	10.5	2.7	10.8	2.6	-0.3	-0.8 to 0.2
Health	12.4	3.2	12.8	3.2	-0.4	-1.0 to 0.2
Role Restriction	17.4	4.8	18.6	4.3	-1.2	-2.1 to -0.4
Depression	17.9	5.2	18.0	4.7	-0.1	-1.0 to 0.8
Spousal Support	16.8	5.0	17.8	5.1	-1.0	-1.9 to -0.1
Child Domain total	94.3	17.6	91.1	14.8	3.1	0.3 to 6.0
Parent Domain total	112.5	22.0	115.7	20.6	-3.2	-7.1 to 0.6
Total Stress	206.8	34.8	207.2	31.7	-0.4	-6.4 to 5.6
Life Stress	10.4	7.4	10.9	8.8	-0.5	-2.0 to 1.0

* n varied slightly by analysis due to missing data.

[†] Adjusted for participants' study site, age at assessment (in months), socioeconomic status (Hollingshead [1975] index, measured continuously), race (white versus other), and gender; 95% CI = 95% confidence interval.

TABLE 3
Fathers' Mean Parenting Stress Index (PSI) Scores for Cases (n = 210^{*}) and Controls (n = 222^{*})

	Cases		Controls		Case-Control Difference [†]	
	Mean	SD	Mean	SD	Difference	95% CI p Value
Child subdomains						
Distractibility	24.8	3.7	25.5	3.5	-0.6	-1.3 to 0.1 .072
Adaptability	25.2	4.8	24.6	4.7	0.6	-0.3 to 1.5 .201
Reinforces Parent	9.1	2.9	9.1	2.7	-0.1	-0.6 to 0.5 .770
Demanding	17.6	3.8	15.8	3.7	1.8	1.1 to 2.5 .000
Mood	9.3	2.8	9.0	2.5	0.3	-0.3 to 0.8 .342
Acceptability	12.4	3.7	11.3	3.0	1.0	0.4 to 1.7 .002
Parent subdomains						
Competence	25.1	5.4	24.6	5.1	0.5	-0.5 to 1.6 .305
Isolation	12.5	3.5	13.0	3.5	-0.5	-1.2 to 0.2 .150
Attachment	12.3	3.1	12.2	2.8	0.0	-0.5 to 0.6 .909
Health	11.5	2.5	11.8	3.2	-0.3	-0.8 to 0.3 .371
Role Restriction	16.6	4.2	17.1	4.2	-0.5	-1.3 to 0.3 .230
Depression	16.4	4.5	16.5	4.2	-0.1	-1.0 to 0.7 .807
Spousal Support	15.0	4.0	15.3	4.1	-0.3	-1.1 to 0.5 .483
Child Domain	98.4	16.3	95.4	15.1	3.0	-0.1 to 6.0 .056
Parent Domain	109.5	20.7	110.5	19.6	-1.0	-4.9 to 2.9 .620
Total Stress	207.9	33.3	206.3	30.8	1.7	-4.5 to 7.8 .591
Life Stress	9.9	7.0	9.4	7.1	0.6	-0.8 to 2.0 .408

* n varied slightly by analysis due to missing data.

[†] Adjusted for participants' study site, age at assessment (in months), socioeconomic status (Hollingshead [1975] index, measured continuously), race (white versus other), and gender; 95% CI = 95% confidence interval.

TABLE 4
Cases' Mean Parenting Stress Index (PSI) Scores for Mothers (n = 246^{*}) and Fathers (n = 210^{*})

	Mothers		Fathers		Mother-Father Difference [†]		
	Mean	SD	Mean	SD	Difference	95% CI	p Value
Child subdomains							
Distractibility	24.7	4.1	24.8	3.7	0.2	-0.4 to 0.7	.584
Adaptability	23.6	5.5	25.2	4.8	1.6	0.9 to 2.3	.000
Reinforces Parent	8.1	2.5	9.1	2.9	1.0	0.5 to 1.4	.000
Demanding	17.5	4.2	17.6	3.8	0.1	-0.5 to 0.7	.762
Mood	8.7	3.1	9.3	2.8	0.6	0.3 to 1.0	.001
Acceptability	11.8	3.7	12.4	3.7	0.5	0.0 to 1.1	.043
Parent subdomains							
Competence	25.2	5.5	25.1	5.4	-0.1	-0.9 to 0.7	.799
Isolation	12.3	4.0	12.5	3.5	0.3	-0.4 to 0.9	.394
Attachment	10.5	2.7	12.3	3.1	1.8	1.3 to 2.2	.000
Health	12.4	3.2	11.5	2.5	-0.8	-1.3 to -0.4	.000
Role Restriction	17.4	4.8	16.6	4.2	-0.8	-1.4 to -0.1	.017
Depression	17.9	5.2	16.4	4.5	-1.5	-2.3 to -0.7	.000
Spousal Support	16.8	5.0	15.0	4.0	-1.8	-2.4 to -1.1	.000
Child Domain	94.3	17.6	98.4	16.3	4.1	1.9 to 6.3	.000
Parent Domain	112.5	22.0	109.5	20.7	-3.0	-6.1 to 0.2	.063
Total Stress	206.7	34.8	207.9	33.3	1.2	-3.6 to 6.0	.629
Life Stress	10.4	7.4	9.9	7.0	-0.5	-1.4 to 0.5	.344

* n varied slightly by analysis due to missing data.

[†] Adjusted for participants' study site, age at assessment (in months), socioeconomic status (Hollingshead [1975] index, measured continuously), race (white versus other), and gender; 95% CI = 95% confidence interval.

TABLE 5
Controls' Mean Parenting Stress Index (PSI) Scores for Mothers (n = 253^{*}) and Fathers (n = 222^{*})

	Mothers		Fathers		Mother-Father Difference [†]		
	Mean	SD	Mean	SD	Difference	95% CI	p Value
Child subdomains							
Distractibility	24.9	4.0	25.5	3.5	0.6	0.0 to 1.2	.047
Adaptability	23.6	5.0	24.6	4.7	1.0	0.3 to 1.7	.005
Reinforces Parent	8.0	2.0	9.1	2.7	1.2	0.8 to 1.6	.000
Demanding	15.5	4.1	15.8	3.7	0.2	-0.4 to 0.8	.433
Mood	8.8	2.6	9.0	2.5	0.3	-0.1 to 0.6	.139
Acceptability	10.3	2.8	11.3	3.0	1.0	0.5 to 1.5	.000
Parent subdomains							
Competence	25.1	5.1	24.6	5.1	-0.5	-1.3 to 0.3	.221
Isolation	12.6	3.5	13.0	3.5	0.4	-0.2 to 1.0	.204
Attachment	10.8	2.6	12.2	2.8	1.5	1.0 to 1.9	.000
Health	12.8	3.2	11.8	3.2	-1.0	-1.5 to -0.4	.000
Role Restriction	18.6	4.3	17.1	4.2	-1.5	-2.2 to -0.9	.000
Depression	18.0	4.7	16.5	4.2	-1.5	-2.3 to -0.8	.000
Spousal Support	17.8	5.1	15.3	4.1	-2.5	-3.2 to -1.9	.000
Child Domain	91.1	14.8	95.4	15.1	4.3	2.1 to 6.5	.000
Parent Domain	115.7	20.6	110.5	19.6	-5.2	-8.3 to -2.2	.001
Total Stress	207.2	31.7	206.2	30.8	-0.9	-5.5 to 3.7	.694
Life Stress	10.9	8.8	9.4	7.1	-1.5	-2.6 to -0.5	.005

* n varied slightly by analysis due to missing data.

[†] Adjusted for participants' study site, age at assessment (in months), socioeconomic status (Hollingshead [1975] index, measured continuously), race (white versus other), and gender; 95% CI = 95% confidence interval.