Published in final edited form as:

Cleft Palate Craniofac J. 2015 November; 52(6): 651–659. doi:10.1597/14-050.

Parent-Reported Family Functioning Among Children With Cleft Lip/Palate

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Abstract

Objective—To examine family functioning related to sociodemographic and clinical characteristics in youth with cleft lip and/or palate (CL/P).

Design—Cross-sectional, multisite investigation.

Setting—Six U.S. cleft centers.

Patients/Participants—A diverse sample of 1200 children with CL/P and their parents.

Main Outcome Measure—Parents completed the Family Environment Scale (FES), which assesses three domains of family functioning: cohesion (or closeness), expressiveness (open expression of feelings), and conflict. Demographic and clinical characteristics were also assessed including race, ethnicity, type of insurance, and surgical recommendations.

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This paper was originally presented at the 12th International Congress on Cleft Lip/Palate and Related Craniofacial Anomalies, Lake Buena Vista, Florida, May 9, 2013.

Results—The FES scores for families seeking team evaluations for their youth with CL/P (mean age = 11.6 years) fall within the average range compared with normative samples. Families receiving surgical recommendations for their youth also had FES scores in the average range, yet families of children recommended for functional surgery reported greater cohesion, expressiveness, and less conflict compared with those recommended for aesthetic surgery (P < .05). For cohesion and expressiveness, significant main effects for race (P = .012, P < .0001, respectively) and ethnicity (P = .004, P < .0001, respectively) were found but not for their interaction. No significant differences were found on the conflict domain. Families with private insurance reported significantly greater cohesion (P < .001) and expressiveness (P < .001) than did families with public insurance.

Conclusions—Family functioning across domains was in the average range. However, observed differences by race, ethnicity, type of insurance, and surgical recommendation may warrant consideration in clinical management for patients and families.

Keywords

adjustment; cleft lip/palate; ethnicity; family functioning; race

It is well established that the diagnosis of a cleft or craniofacial condition affects not only the affected child but the family as well (Endriga and Kapp-Simon, 1999; Pope, 1999; Collett and Speltz, 2007; Baker et al., 2009). Adjustment to a chronic condition such as cleft lip and/or palate (CL/P) is thought to be associated with numerous factors, including condition-specific medical factors (e.g., type and severity of cleft diagnosis, visibility of the condition, surgical interventions); social-ecological factors, such as economic resources, access to care, and social support (Wallander and Varni, 1998); demographic variables, including race, ethnicity, and gender (Berger and Dalton, 2011); and child and family characteristics, such as self-concept, coping style, and family adjustment (Lavigne and Faier-Routman, 1993; Barlow and Ellard, 2006). The complex interrelationships among such variables are not well understood.

Family functioning is a construct that is receiving increased empirical attention in relation to adjustment to chronic health conditions (Hanson et al., 1992; Drotar, 1997; McClellan and Cohen, 2007; Herzer et al., 2010). Family characteristics, such as degree of cohesion or closeness, have been identified as risk and protective factors for adjustment within the broader pediatric illness and child development literatures (McClellan and Cohen, 2007; Herzer et al., 2010). Greater family cohesion and expressiveness (or ability to express feelings directly and to talk about personal problems) and less conflict generally are associated with self-confidence, social competence, and psychological health among youth and young adults (Oliver and Paull, 1995; Ketsetzis et al., 1998). Among children with chronic conditions and their families, high levels of cohesion are associated with positive health outcomes (Drotar, 1997; Newby, 2000). Further, good family functioning has been associated with positive quality of life among youth with chronic medical conditions (Herzer et al., 2010). In contrast, high levels of conflict are frequently associated with emotional and behavior problems in children with chronic conditions (Drotar, 1997).

In the broader pediatric chronic illness literature, family functioning has been shown to be important in terms of adaptation to stress for children and parents; during times of stress, strong cohesion and expressiveness have been associated with positive child adjustment (Holmes et al., 1999; Jobe-Shields et al., 2009). Families having children with CL/P are vulnerable to ongoing stress related to their child's condition given the need for surgical, dental, speech, and other types of interventions, all of which can impose significant burdens for families, especially for those with limited financial and health care resources such as insurance. The experience of having a child with a facial difference may also be conceptualized as an ongoing stressor, given that these children may be vulnerable to peer victimization and other forms of social stigmatization (Strauss et al., 2007).

Despite these known stressors, there are scant data available regarding family functioning among youth with CL/P and whether there are differences compared with normative family systems, that is, compared with those families having children who do not have a chronic health condition such as CL/P. Furthermore, there has been limited investigation of how current treatment needs (e.g., recommendation for additional surgery) and sociocultural factors such as race, ethnicity, and insurance status are associated with family functioning and, more broadly, quality of life among children with CL/P. Data from our ongoing longitudinal study of youth with CL/P reveal that current surgical treatment needs are associated with reduced quality of life, and health insurance status and ethnicity/race are risk factors identified as part of this model (Broder et al., 2014). Families of children with surgical needs may be experiencing additional stressors (e.g., social stigmatization related to their children's visible differences, time and financial burdens associated with upcoming surgery) that may affect family functioning.

In addition, race and ethnicity are associated with family functioning; greater levels of cohesion and expressiveness have been reported among ethnic minority families (McEachern and Kenny, 2002; Moos and Moos, 2002; Clay et al., 2007). For example, greater levels of cohesion have been associated with lower levels of psychological distress among U.S. Latinos (Rivera et al., 2008). Among Asian American adolescents, greater levels of conflict and lower levels of expressiveness have been reported compared with adolescents of European descent (Greenberger and Chen, 1996). However, relationships between race, ethnicity, and family functioning have yet to be studied among families of children with CLP. Potential differences in family functioning among youth of varying racial or ethnic backgrounds may have important implications for understanding how families adjust to their child's condition and cope over time. For example, greater cohesion and expressiveness, which have been associated with more positive adjustment among youth with chronic conditions (Holmes et al., 1999; Jobe-Shields et al., 2009) and among ethnic minority families (McEachern and Kenny, 2002) may serve as protective factors and contribute to positive adjustment. Alternately, higher levels of family conflict may be more prevalent among certain racial/ethnic groups and may constitute a risk factor for problematic adjustment or coping. Finally, racial and ethnic differences in family functioning are important to understand given that there are known racial and ethnic disparities in healthrelated quality of life and overall health (U.S. Department of Health and Human Services, 2000; Institute of Medicine, 2003; Flores and Tomany-Korman, 2008), and contextual differences within families may contribute to these disparities (Wallander et al., 2012).

To our knowledge, only one study to date has directly assessed family functioning using the Family Environment Scale (FES; Moos and Moos, 2002) in a sample of 165 children (mean age = 6.4 years) with CL/P in Argentina (Wyszynski et al., 2005). Compared with a control group, families of children with CL/P scored higher on cohesion and showed lower levels of conflict and above-average expressiveness. Nevertheless, family functioning has yet to be assessed in samples of U.S. youth with CL/P, and family functioning has not been evaluated in families of youth with CL/P in relationship to surgical needs, race/ethnicity, or insurance status.

This study examined the following research questions: (1) Does family functioning among youth with CL/P differ compared with normative data from families of children without cleft or chronic health conditions? (2) Does family functioning among youth with CL/P differ based on youths' current surgical treatment needs? (3) Are there any differences in family functioning across sociocultural variables such as race, ethnicity, and type of insurance coverage?

Method

Participants

A sample of 1200 parent-child dyads at six U.S. cleft centers (Children's Healthcare of Atlanta, The Children's Hospital of Philadelphia, Lancaster Cleft Palate Clinic, New York University, University of Illinois at Chicago, and University of North Carolina—Chapel Hill) was recruited as part of our ongoing longitudinal, observational investigation of quality of life among youth with CL/P. These centers represent rural and urban locations with diverse ethnic/racial groups. Institutional review board approval was granted at all sites, and informed consent/assent was obtained from all participants. English and/or Spanish-speaking children with CL/P between the ages of 7.5 and 18 years and their caregivers were eligible to participate. This age range was selected because the primary aim of the larger study was to examine quality of life among youth undergoing secondary functional and/or aesthetic surgical procedures such as bone grafting and scar revision, which are typically performed during middle childhood and adolescence. Exclusion criteria included inability to read at a second-grade level, diagnosis with an incomplete cleft lip without cleft of the alveolus, or diagnosis with a craniofacial syndrome (e.g., Apert syndrome) or other complex medical conditions (e.g., heart problems).

Procedure

Families were recruited and data collection was completed at the time of the child's regularly scheduled clinic visit with the cleft team or individual cleft team providers. Parents and their children were asked to complete questionnaires independently. Spanish versions of all questionnaires were used for Spanish-speaking parents and/or parent-child dyads and interpreters were available, as needed, at all sites. Surgeons also completed a rating form (described below) at the time of the child's visit regarding current surgical needs. Data presented in this report were obtained from the baseline assessment, and analyses are based on parent completed measures described below as well as clinical recommendations from the sites' surgeons.

Measures

Brief Demographic Questionnaire—This questionnaire included information about the child's gender, age, race, academic grade, and ethnicity. The categories for race and ethnicity used in the present study are based on National Institutes of Health standards for maintaining, collecting, and presenting data about race (American Indian or Alaskan Native, Asian or Pacific Islander, black, white, or more than one race) and ethnicity (Hispanic or Latino origin and not of Hispanic or Latino origin). The caregiver was also asked to indicate payer source (e.g., private insurance, Medicaid, self-pay) as one indicator of the family's financial resources.

Family Environment Scale—The FES (Moos and Moos, 2002) is composed of 90 truefalse items measuring social and environmental characteristics of families. The scale is based on a three-dimensional conceptualization of families. The Relationship dimension has been chosen for this study because of its relevance to social functioning risk factors in populations with chronic conditions (Newby et al., 2000) and personal correspondence with the developers of the FES. It consists of 27 items that assess three domains: family cohesion, or the degree of commitment, help, and support family members provide for each other (e.g., "There is a feeling of togetherness in our family"); expressiveness, or the extent to which family members are encouraged to express their feelings directly and to talk about personal problems (e.g., "Family members often keep their feelings to themselves"); and conflict, or the amount of openly expressed anger, criticism, and fighting among family members (e.g., "We fight a lot in our family"). Raw scores can range from 0 to 9 on each subscale; higher scores reflect greater cohesion/expressiveness/conflict. Raw scores are converted to standard scores using instructions provided in appendix A of the FES manual (Moos and Moos, 2002). This instrument has moderate to high internal consistencies and acceptable test-retest reliabilities (Moos and Moos, 2002). It has been widely used in studies with families of children with chronic conditions to examine how the family affects the course of treatment and psychosocial outcome of the condition (Alderfer et al., 2008).

Surgical Recommendations—At the time of the child's cleft-related office visit, the treating surgeon completed a clinical rating form indicating surgical recommendation and procedure, if any, within 1 year. For the purposes of this study, recommended reconstructive surgical procedures were categorized as either functional (e.g., cleft palate revision, bone grafting) or aesthetic (e.g., tip rhinoplasty, scar revision).

Statistical Analyses

Descriptive statistics (means, frequencies) were calculated. Raw scores for the FES domains were converted into standard scores. FES domain scores were assessed using multivariate analysis of variance (MANOVA) via generalized linear model to examine differences for race, ethnicity, and their interaction. Tukey's HSD post hoc analyses were performed to detect differences among ethnic and racial groups. *t* Tests were used to examine differences for type of insurance (private versus non-private payer) and surgical recommendations: aesthetic (e.g., scar revision, tip rhinoplasty) versus functional (e.g., palatal revision).

Results

The mean age for children was 11.6 ± 3.1 years. More than half of the sample (56%) identified as male. Seventy-six percent had a diagnosis of cleft lip and palate, and 24% had cleft palate only. With respect to ethnicity, 16% of the sample identified as Hispanic or Latino. Most of the sample identified as Caucasian (68%), 12% as other, 11% as African American, and 10% as Asian. More than half of the sample (55%) reported having private insurance. Thirty-six percent of the sample (n = 433) received a recommendation for secondary surgery at their baseline visit. Of these, 225 (52%) received recommendations for aesthetic surgery and 208 (48%) received a recommendation for functional surgery.

As shown in Table 1, scores on the FES domains for the total sample fell within the normal range (standard score of 50 with standard deviation of 10). Normative data for the FES are derived from a large, representative sample (n = 1432), which included families of all age groups, racial and ethnic minority groups, and family structure (e.g., single parent, multigenerational families; Moos and Moos, 2002). Compared with this normative sample, cohesion and expressiveness domain raw scores were higher and conflict domain raw scores were lower among this sample of families of children with CLP; however, all standard scores for the CL/P sample fall within one standard deviation of the normative samples' scores.

Surgical Recommendation and FES Domains

Standard scores for the three FES domains were compared via independent *t* test for youth who had been referred for any type of secondary surgery versus those who had not received a surgical recommendation. No significant differences were found between groups for cohesion, expressiveness, or conflict.

Standard scores for the cohesion, expressiveness, and conflict domains of the FES were then compared for children who had been referred for aesthetic surgery (e.g., scar revision, tip rhinoplasty) versus functional surgery (e.g., bone grafting or palate revision; see Table 2). Results of the independent t test analysis indicated that for cohesion, there was a trend for higher scores in the functional group, t(410) = 1.94, P = .053, suggesting a greater sense of closeness among these families compared with families of children referred for aesthetic procedures.

For the expressiveness domain, scores were significantly higher in the functional group, suggesting greater parent-rated expressiveness, t(397) = -2.34, P = .020. Lastly, for the conflict domain, significantly lower scores were reported for the functional group compared with the aesthetic group, suggesting less parent-reported conflict, t(403) = 3.00, P = .003.

Race and Ethnicity

All of the scores for the FES domains by race and ethnicity fell within broad limits of the average for standard scores. As shown in Table 3, the MANOVA analyses revealed significant main effects for race and ethnicity but not for their interaction. Significant main effects for race (P < .012) and ethnicity (P = .004) were found on the cohesion domain and for ethnicity (P < .0001) and race (P < .0001) on the expressiveness domain but not for their

interaction (P = .18 and P = .39 for cohesion and expressiveness, respectively; see Table 4). There were no significant main effects or interactions for race and ethnicity on the conflict scale. Specifically, on the cohesion domain, non-Hispanic families reported significantly greater cohesion compared with families identifying as Hispanic or Latino. Tukey's HSD post hoc analyses indicated that for race, African American families scored significantly higher for cohesion compared with Asian families (55.70 versus 52.42, P < .05), and Caucasian families scored significantly higher on cohesion compared with African American families (58.15 versus 55.70, P < .05; see Table 5).

On the expressiveness domain, non-Hispanic families scored significantly higher than the Hispanic or Latino families (54.83 versus 50.07, P = .029). As shown in Table 6, Tukey's HSD post hoc analyses revealed that Caucasian families scored significantly higher on expressiveness compared with both African American and other families (53.13 versus 50.14 versus 51.76, respectively; P < .05). Asian Americans also scored significantly lower on this domain than the African American and other families (47.44 versus 50.14 versus 51.76, respectively, P < .05).

Type of Insurance

Lastly, we examined differences in the domain scores by type of insurance (see Table 7). Independent t test analyses revealed that there was no significant difference between groups on the conflict domain (P = .394). However, those with private insurance reported significantly greater expressiveness, t(1090) = 6.22, P < .001, and cohesion, t(1106) = 3.88, P < .001, compared with those with public insurance.

Discussion

This study examined family functioning using the FES in a sample of 1200 children with CL/P and their parents who were seeking evaluation from an established cleft team. Specifically, we were interested in exploring whether there were differences related to the presence of treatment needs and specific secondary surgery recommendations (functional versus aesthetic). We also aimed to determine whether there were differences in family functioning related to race and ethnicity, since these variables are known to be associated with differences in family functioning (Moos and Moos, 2002; McEachern and Kenny, 2002; Clay et al., 2007). Finally, we examined differences related to payer type (private versus public insurance) given that families with fewer financial resources may be differentially affected by the cleft diagnosis and its multiple associated treatments and surgeries (Abbott et al., 2011; Broder et al., 2012).

Overall, family functioning scores for this sample of youth with CL/P are within the normal range and are suggestive of healthy levels of cohesion, expressiveness, and conflict. However, our findings suggest that sociocultural variables such as race, ethnicity, type of insurance, and clinical need are related to domains of family functioning. Given the salience of self-image and quality of life in cleft outcomes, attention to family issues is crucial. While unmet surgical needs are linked to youths' quality of life, unmet surgical needs are also linked to health disparities in racial/ethnic minorities and in relation to having nonprivate insurance (Abbott et al., 2011; Broder et al., 2012). Furthermore, our findings support prior

research that has found associations between adjustment among parents and children with cleft conditions. For example, a recent study found that parental coping, adolescent facial appearance, and psychological adjustment were linked (Berger and Dalton, 2011). Similarly, Murray and colleagues (2010) found that poorer parenting environments (e.g., less cognitive stimulation and emotional support) were associated with increased risk for socioemotional problems in school-aged children with CL/P.

Our results are also consistent with those of Wyszynski and colleagues (2005), who examined family functioning domains in a sample of Argentinian families of children with orofacial clefts (mean age = 6.4 years). The raw scores from our study regarding cohesion, expressiveness, and conflict are nearly identical to Wyszynski's sample, thereby suggesting that our families are also reporting high levels of cohesion and expressiveness and low levels of conflict on average. Both samples included school-age children; thus, it appears that at this stage, most families are functioning well despite their child's condition. However, longitudinal analyses are indicated to better understand how family functioning changes over time and whether there are time periods (e.g., infancy) when condition and treatment-related stressors may be more salient and have greater effects on family functioning.

Interestingly, we found that families identifying as non-Hispanic or Latino and/or Caucasian reported greater cohesion and expressiveness compared with African American and Latino families. Our results are somewhat contradictory to the broader literature whereby African American and Latino families tend to be more cohesive and expressive than Caucasian families (McEachern and Kenny, 2002; Negy and Snyder, 2006; Clay et al., 2007). Furthermore, we observed that Asian American families reported significantly less expressiveness compared with families identifying as African American or other. Although statistical significance was not reached (perhaps because of our small Asian American sample size), adjusted means are also suggestive of lower expressiveness in Asian American families compared with Caucasian families. In the broader literature, Asian American families have been described as less cohesive and expressive and higher in conflict than their European American counterparts (Greenberger and Chen, 1996). While there are vast differences within ethnic groups, several factors may account for the differences observed in our study and warrant consideration. For example, issues related to "saving face" (impression management) and reluctance to disclose personal health information, particularly among Asian families, may also contribute to the differences on the FES observed in our sample. Level of acculturation may negatively affect cohesiveness among Latino families (Rivera et al., 2008). Furthermore, most studies that have examined dimensions of family functioning among persons from varying racial and ethnic backgrounds have not been conducted in populations who are also coping with a chronic and potentially stigmatizing condition such as CL/P. In short, additional research is needed to better understand family functioning among ethnic minority families of youth with CL/P.

Overall, families in our sample reported low levels of conflict. However, there was an interesting trend between families of children recommended for functional versus aesthetic surgery, with greater conflict noted in the latter group. Although the FES does not specifically assess conflict about surgery, it could be that the higher conflict scores may

reflect disagreements between parents and youth about the need for additional aesthetic procedures and/or tension associated with the social stresses on youth and their families around issues related to social anxiety and coping with their facial differences. Clinically, such disagreements are reportedly not uncommon, particularly when children reach adolescence (Kapp-Simon, 1995). This finding emphasizes the importance of obtaining input from both youth and parents around aesthetic surgical decision making.

Differences in family functioning were also observed by type of insurance; specifically, greater cohesion and expressiveness were reported among those with private insurance compared with those with public insurance. These differences may reflect known racial disparities in health care access and coverage in that racial and ethnic minorities are more likely to have public insurance (Flores and Tomany-Korman, 2008), and in our sample, families identifying as Caucasian or non-Hispanic or Latino reported greater cohesion and expressiveness compared with those from other racial/ethnic groups. Families with private insurance may be able to more easily adhere to recommended surgeries and interventions for their cleft-affected child, which in turn may enhance well-being and quality of life (Broder et al., 2012).

Clinical Implications

Family functioning, particularly expressiveness and cohesion, contributes to psychological well-being and adjustment in cleft populations and is therefore important to assess clinically. Family functioning is a powerful determinant of overall quality of life and well-being in youth with chronic medical issues (Newby, 2000). Specifically, supportive family relationships are linked to positive psychological functioning (Hanson et al., 1992; Grey et al., 1998; Broder et al., 2014). These issues are particularly salient when life stressors such as surgery and hospitalization are imminent. Research with families of children with chronic conditions underscores the importance of positive coping skills and adequate social support (Baker et al., 2009; Berger and Dalton, 2011).

Cleft habilitation (e.g., appointments, hospitalizations) can impose a significant burden of care on families, including missing days of work, obtaining childcare for siblings of the affected children, and requiring health care resources such as insurance coverage. These stressors can weigh heavily on the family system. As part of the American Cleft Palate Craniofacial Association's Parameters for Evaluation and Treatment of Patients With Cleft Lip/Palate or Other Craniofacial Anomalies (2009), routine psychological evaluations are recommended, which generally focus on the identified patient. However, family coping and associated psychosocial issues may be particularly salient among families with poor cohesion, single parents with limited social support, and families in which there is significant conflict. Currently, estimates indicate that low family support and single parents exist in 30% to 40% of families, and this number is higher in systems having a child with a chronic health condition (Brown et al., 2008). Our findings underscore the need to carefully consider family functioning and other contextual factors such as culture and socioeconomic resources in these assessments to better understand how these factors affect decision making around surgery and other treatments and to identify families who may benefit from psychosocial intervention and/or support.

Given that episodic surgical and other interventions occur in cleft care, it is suggested that screenings include assessment of families' coping and support, particularly for those with low levels of cohesion or high levels conflict. Involvement of the family and the patient in decision making is critical as such involvement is reportedly associated with positive outcomes (Sloper, 2000; Barlow and Ellard, 2006). Further, direct interventions aimed at parents of children with chronic conditions (e.g., cleft, Down syndrome) have been shown to have positive impacts on family functioning (Pelchat et al., 1999), which is congruent with a recent systematic review of qualitative and quantitative studies on family function (Cousino and Hazen, 2013).

Limitations and Future Directions

Although this data set represents the largest study of youth with clefts in the United States, the data presented are cross-sectional and are able to identify relationships between family functioning and sociodemographic and clinical variables at only one point in time. We also did not have a control group for this study, although the FES is a normed instrument, which enabled us to examine family functioning domains among youth with CL/P in relation to normative data. Longitudinal analyses that use appropriate controls or comparison groups are indicated to understand whether issues regarding family functioning change over time and/or represent risk or protective factors regarding the youths' clinical needs and emerging stressors (e.g., surgery). Further, we are likely overpowered because of the large sample, which reveals significance that may not be clinically meaningful (e.g., differences between racial groups, ethnicity). Nonetheless, race and ethnicity are critical components of research related to health utilization and health outcomes (Broder et al., 2012) and warrant further study in this population. In examining issues related to sociocultural differences such as ethnicity, length of time since establishing residency in the United States, degree of acculturation, educational level, and socioeconomic status may be relevant components to consider in examining family functioning and perceived burden of care. In addition, it may be important to examine youths' perceptions of family functioning, the quality of the family's support system, and parents' and youths' specific coping skills. This recommendation is consistent with findings reported by Baker et al. (2009). Examination of siblings and their perceptions of family functioning may also be relevant as their coping and adjustment are additional risk and protective factors that can affect the family system (Sharpe and Rossiter, 2002; Brennan et al., 2013).

Our findings should be replicated, and qualitative data on family functioning may provide essential information related to health beliefs, specific conflict areas, as well as facilitators of positive family function. Stressors such as the experience of having a child with CL/P can be viewed as opportunities for families to bolster competencies, particularly in the presence of protective factors (e.g., low conflict, healthy levels of cohesion and expressiveness), which in turn may enhance resilience and well-being (Fisman et al., 1996; Gomez and McLaren, 2006). Additional research could provide critical detail regarding the nature of the burdens that affect families as well as adaptive skills and coping styles to foster resilience in the system. Such research could also provide more information on cohesion and psychological hardiness within varied sociocultural contexts, particularly when medical, financial, and social factors are present. In summary, our findings support prior research and

clinical recommendations to incorporate family adjustment assessment in our team care for patients and their families (Pelchat et al., 1999; Baker et al., 2009; Berger and Dalton, 2011).

Acknowledgments

We would like to thank the caregivers and their children who took part in this study as well as clinical personnel from the participating sites. We would also like to acknowledge Dr. Robert Norman for his assistance with statistical analyses.

This research was supported by the National Institutes of Health, National Institute of Dental and Craniofacial Research (grant DE018729 to H. Broder, principal investigator, and grant K23 DE020854 to C. Crerand, principal investigator).

References

- Abbott MM, Kokorowski PJ, Meara JG. Timeliness of surgical care in children with special health care needs: delayed palate repair for publicly insured and minority children with cleft palate. J Pediatr Surg. 2011; 46:1319–1324. [PubMed: 21763828]
- Alderfer MA, Fiese BH, Gold JI, Cutuli JJ, Holmbeck GN, Goldbeck L, Chambers CT, Abad M, Spetter D, Patterson J. Evidence-based assessment in pediatric psychology: family measures. J Pediatr Psychol. 2008; 33:1046–1061. [PubMed: 17905801]
- American Cleft Palate Craniofacial Association. [Accessed January 14, 2014] Parameters for Evaluation and Treatment of Patients With Cleft Lip/Palate or Other Craniofacial Anomalies—Revision. 2009. Available at http://www.acpa-cpf.org/uploads/site/Parameters_Rev_2009.pdf
- Baker SR, Owens J, Stern M, Willmot D. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. Cleft Palate Craniofac J. 2009; 46:229–236. [PubMed: 19642758]
- Barlow JH, Ellard DR. The psychosocial well-being of children with chronic disease, their parents and siblings: an overview of the research evidence base. Child Care Health Dev. 2006; 32:19–31. [PubMed: 16398788]
- Berger ZE, Dalton LJ. Coping with a cleft II: factors associated with psychosocial adjustment of adolescents with a cleft lip and palate and their parents. Cleft Palate Craniofac J. 2011; 48:82–90. [PubMed: 20500070]
- Brennan C, Hugh-Jones S, Aldridge J. Paediatric life-limiting conditions: coping and adjustment in siblings. J Health Psychol. 2013; 18:813–824. [PubMed: 22947892]
- Broder HL, Wilson-Genderson M, Sischo L. Examination of a theoretical model for oral health-related quality of life among youths with cleft. Am J Public Health. 2014; 104:865–871. [PubMed: 24625170]
- Broder HL, Wilson-Genderson M, Sischo L. Health disparities among children with cleft. Am J Public Health. 2012; 102:828–830. [PubMed: 22420792]
- Brown RT, Wiener L, Kupst MJ, Brennan T, Behrman R, Compas BE, David Elkin T, Fairclough DL, Friebert S, Katz E, et al. Single parents of children with chronic illness: an understudied phenomenon. J Pediatr Psychol. 2008; 33:408–421. [PubMed: 17906331]
- Clay CM, Ellis MA, Griffin ML, Amodeo M, Fassler IR. Black women and white women: do perceptions of childhood family environment differ? Fam Process. 2007; 46:243–256. [PubMed: 17593888]
- Collett BR, Speltz ML. A developmental approach to mental health for children and adolescents with orofacial clefts. Orthod Craniofac Res. 2007; 10:138–148. [PubMed: 17651130]
- Cousino MK, Hazen RA. Parenting stress among caregivers of children with chronic illness: a systematic review. J Pediatr Psychol. 2013; 38:809–828. [PubMed: 23843630]
- Drotar D. Relating parent and family functioning to the psychological adjustment of children with chronic health conditions: what have we learned? What do we need to know? J Pediatr Psychol. 1997; 22:149–165. [PubMed: 9114640]

Endriga MC, Kapp-Simon KA. Psychological issues in craniofacial care: state of the art. Cleft Palate Craniofac J. 1999; 36:3–11. [PubMed: 10067755]

- Fisman S, Wolf L, Ellison D, Gillis B, Freeman T, Szatmari P. Risk and protective factors affecting the adjustment of siblings of children with chronic disabilities. J Am Acad Child Adolesc Psychiatry. 1996; 35:1532–1541. [PubMed: 8936921]
- Flores G, Tomany-Korman SC. Racial and ethnic disparities in medical and dental health, access to care, and use of services in US children. Pediatrics. 2008; 121:e286–e298. [PubMed: 18195000]
- Gomez R, McLaren S. The association of avoidance coping style, and perceived mother and father support with anxiety/depression among late adolescents: applicability of resiliency models. Personality and Individual Differences. 2006; 40:1165–1176.
- Greenberger E, Chen C. Perceived family relationships and depressed mood in early and late adolescence: a comparison of European and Asian Americans. Dev Psychol. 1996; 32:707–716.
- Grey M, Boland EA, Yu C, Sullivan-Bolyai S, Tamborlane WV. Personal and family factors associated with quality of life in adolescents with diabetes. Diabetes Care. 1998; 21:909–914. [PubMed: 9614606]
- Hanson CL, De Guire MJ, Schinkel AM, Henggeler SW, Burghen GA. Comparing social learning and family systems correlates of adaptation in youths with IDDM. J Pediatr Psychol. 1992; 17:555–572. [PubMed: 1432481]
- Herzer M, Godiwala N, Hommel KA, Driscoll K, Mitchell M, Crosby LE, Piazza-Waggoner C, Zeller MH, Modi AC. Family functioning in the context of pediatric chronic conditions. J Dev Behav Pediatr. 2010; 31:26–34. [PubMed: 20081433]
- Holmes CS, Yu Z, Frentz J. Chronic and discrete stress as predictors of children's adjustment. J Consult Clin Psychol. 1999; 67:411–419. [PubMed: 10369062]
- Institute of Medicine. Unequal Treatment: Confronting Racial and Ethnic Disparities in Health Care. National Academies Press; Washington, DC: 2003.
- Jobe-Shields L, Alderfer MA, Barrera M, Vannatta K, Currier JM, Phipps S. Parental depression and family environment predict distress in children before stem cell transplantation. J Dev Behav Pediatr. 2009; 30:140–146. [PubMed: 19322103]
- Kapp-Simon KA. Psychological interventions for the adolescent with cleft lip and palate. Cleft Palate Craniofac J. 1995; 32:104–108. [PubMed: 7748869]
- Ketsetzis M, Ryan BA, Adams GR. Family processes, parent-child interactions, and child characteristics influencing school-based social adjustment. Journal of Marriage and Family. 1998; 60:374–387.
- Lavigne JV, Faier-Routman J. Correlates of psychological adjustment to pediatric physical disorders: a meta-analytic review and comparison with existing models. J Dev Behav Pediatr. 1993; 14:117–123. [PubMed: 8473527]
- McClellan CB, Cohen LL. Family functioning in children with chronic illness compared with healthy controls: a critical review. J Pediatr. 2007; 150:221–223. [PubMed: 17307532]
- McEachern AG, Kenny MC. A comparison of family environment characteristics among white (non-Hispanic), Hispanic, and African Caribbean groups. Journal of Multicultural Counseling and Development. 2002; 30:40–58.
- Moos, RH.; Moos, BS. Family Environment Scale Manual. 3rd ed.. Mind Garden; Menlo Park, CA: 2002.
- Murray L, Arteche A, Bingley C, Hentges F, Bishop DV, Dalton L, Goodacre T, Hill J. Cleft Lip and Palate Study Team. The effect of cleft lip on socio-emotional functioning in school-aged children. J Child Psychol Psychiatry. 2010; 51:94–103. [PubMed: 19968739]
- Negy C, Snyder DK. Assessing family-of-origin functioning in Mexican American adults: retrospective application of the Family Environment Scale. Assessment. 2006; 13:396–405. [PubMed: 17050910]
- Newby WL, Brown RT, Pawletko TM, Gold SH, Whitt K. Social skills and psychological adjustment of child and adolescent cancer survivors. Psycho-oncology. 2000; 9:113–126. [PubMed: 10767749]
- Oliver JM, Paull JC. Self-esteem and self-efficacy; perceived parenting and family climate; and depression in university students. J Clin Psychol. 1995; 51:467–481. [PubMed: 7593666]

Pelchat D, Bisson J, Ricard N, Perreault M, Bouchard JM. Longitudinal effects of an early family intervention programme on the adaptation of parents of children with a disability. Int J Nurs Stud. 1999; 36:465–477. [PubMed: 10576117]

- Pope AW. Points of risk and opportunity for parents of children with craniofacial conditions. Cleft Palate Craniofac J. 1999; 36:36–39. [PubMed: 10067759]
- Rivera FI, Guarnaccia PJ, Mulvaney-Day N, Lin JY, Torres M, Alegria M. Family cohesion and its relationship to psychological distress among Latino groups. Hisp J Behav Sci. 2008; 30:357–378. [PubMed: 19444326]
- Sharpe D, Rossiter L. Siblings of children with a chronic illness: a meta-analysis. J Pediatr Psychol. 2002; 27:699–710. [PubMed: 12403860]
- Sloper P. Predictors of distress in parents of children with cancer: a prospective study. J Pediatr Psychol. 2000; 25:79–91. [PubMed: 10820946]
- Strauss RP, Ramsey BL, Edwards TC, Topolski TD, Kapp-Simon KA, Thomas CR, Fenson C, Patrick DL. Stigma experiences in youth with facial differences: a multi-site study of adolescents and their mothers. Orthod Craniofac Res. 2007; 10:96–103. [PubMed: 17552946]
- U.S. Department of Health and Human Services. Oral Health in America: A Report of the Surgeon General—Executive Summary. U.S. Department of Health and Human Services; Washington, DC: 2000
- Wallander JL, Fradkin C, Chien AT, Mrug S, Banspach SW, Davies S, Elliott MN, Franzini L, Schuster MA. Racial/ethnic disparities in health-related quality of life and health in children are largely mediated by family contextual differences. Acad Pediatr. 2012; 12:532–538. [PubMed: 22884796]
- Wallander JL, Varni JW. Effects of pediatric chronic physical disorders on child and family adjustment. J Child Psychol Psychiatry. 1998; 39:29–46. [PubMed: 9534085]
- Wyszynski DF, Perandones C, Yannibelli P, Bennun RD. Family environment of individuals with oral clefts in Argentina. Cleft Palate Craniofac J. 2005; 42:185–191. [PubMed: 15748110]

TABLE 1
Family Environment Scale (FES) Domain Scores (Mean, SD) for CL/P Sample (by Race and Ethnicity) and Normative Samples

CL/P Sample	Cohesion	Expressiveness	Conflict
Total (n = 1200)			
Raw	8.00 ± 1.31	6.20 ± 1.67	2.20 ± 1.80
Standard	58.57 ± 8.79	54.07 ± 10.26	44.91 ± 9.49
Race			
Caucasian	58.99 ± 8.66	55.10 610.44	44.55 ± 9.59
African American	56.28 ± 10.84	50.04 ± 8.36	45.86 ± 9.57
Asian	59.34 ± 7.76	54.52 ± 9.98	46.35 ± 9.54
Other	57.23 ± 8.18	50.41 ± 9.30	44.87 ± 8.63
Ethnicity			
Hispanic	56.56 ± 9.32	49.90 ± 9.10	45.53 ± 8.88
Non-Hispanic	59.00 ± 8.62	54.84 ± 10.29	44.76 ± 9.62
FES norms			
Normal family sample (n = 1432)*	6.73 ± 1.47	5.54 ± 1.61	3.18 ± 1.91
African American and Latino families (n = 454)*	6.90 ± 1.94	4.97 ± 1.73	3.26 ± 2.12

^{*} Moos RH, Moos BS. Family Environment Scale Manual. 4th ed. Menlo Park, CA: Mind Garden; 2009.

TABLE 2 Family Functioning Domains by Surgical Recommendation and Type of Surgery

	Surgery Recommended	No Surgery Recommended				Aesthetic	Functional			
FES Domain	(n = 433)	$(n=766)^*$	t	df	Ь	(n = 225)	(n = 208)	t	df	Ь
Cohesion	57.92 ± 9.32	58.92 ± 8.47 1.86 1146 .063	1.86	1146	.063	57.06 ± 10.15	57.06 ± 10.15 58.84 ± 8.28 -1.94 410 $.053$	-1.94	410	.053
Expressiveness	53.80 ± 9.95	54.19 ± 10.42	.61	1129	.543	52.69 ± 10.29	55.01 ± 9.46	-2.34	397	.020
Conflict	44.97 ± 9.86	44.88 ± 9.29	.16 1139	1139	.876	46.35 ± 10.08	43.43 ± 9.41	3.00	403	.003

*
Surgery recommendation was unknown for one participant; the total reported here is n = 1199 for surgery recommendation (yes versus no). FES = Family Environment Scale.

TABLE 3
Multivariate Analysis of Variance Results for Family Environment Scale (FES) Domains

FES Domain	df	Sum of Squares	Mean Square	F	P
Cohesion					
Model	7	1815.98	259.43	3.44	.001
Error	1114	84,127.52	75.52		
Expressiveness					
Model	7	5596.32	799.48	7.88	.0001
Error	1098	111,457.71	101.51		
Conflict					
Model	7	553.24	79.03	0.87	.527
Error	1111	100,574.29	90.53		

Crerand et al. Page 17

TABLE 4
Multivariate Analysis of Variance: Family Environment Scale Domains by Race and Ethnicity

Domain	df	Sum of Squares	Mean Square	F	P
Cohesion					
Race	3	825.09	275.031	3.64	.012
Ethnicity	1	624.12	624.12	8.26	.004
Interaction	3	366.77	122.26	1.62	.183
Expressiveness					
Race	3	3563.65	1187.88	11.7	<.0001
Ethnicity	1	1725.83	1725.83	17.0	<.0001
Interaction	3	306.84	102.28	1.01	.389
Conflict					
Race	3	393.34	131.11	1.45	.227
Ethnicity	1	154.68	154.68	1.71	.191
Interaction	3	5.22	1.74	0.02	.996

TABLE 5
Post Hoc Analyses: Family Environment Scale Cohesion Domain Scores by Race

Race (I)	Race (J)	Mean Difference (I - J)	95% Confidence Interval
Asian	Caucasian	0.69	-1.51, 2.89
	Other	1.64	-1.24, 4.52
	African American*	3.37	0.36, 6.37
Caucasian	Asian	-0.69	-2.89, 1.51
	Other	0.95	-1.23,3.13
	African American*	2.68	0.34, 5.01
Other	Asian	-1.64	-4.52, 1.24
	African American	1.73	-1.26, 4.71
	Caucasian	-0.95	-3.13, 1.23
African American	Asian*	-3.37	-6.37, -0.36
	Caucasian*	-2.68	-5.01, -0.34
	Other	-1.73	-4.71, 1.26

^{*} Tukey's post hoc values are reported, *P < .05.

TABLE 6
Post Hoc Analyses: Family Environment Scale Expressiveness Domain Scores by Race

Race (I)	Race (J)	Mean Difference (I - J)	95% Confidence Interval
Asian	Caucasian	0.29	-2.85, 2.26
	Other*	3.93	0.55, 7.31
	African American*	4.65	1.12, 8.17
Caucasian	Asian	0.29	-2.26, 2.85
	Other*	4.22	1.64, 6.80
	African American*	4.94	2.17, 7.71
Other	Asian*	-3.93	-7.31, -0.55
	African American	0.72	-2.83, 4.26
	Caucasian*	-4.22	-6.80, -1.64
African American	Asian*	-4.65	-8.17, -1.12
	Caucasian*	-4.94	-7.71, -2.17
	Other	-0.72	-4.26, 2.83

^{*} Tukey's post hoc values are reported, *P< .05.

TABLE 7
Family Functioning Domains Means Test by Type of Insurance

	Private Insurance	Non-private insurance	t	df	P
Cohesion	59.57 ± 8.53	57.55 ± 8.61	3.88	1106	<.001
Expressiveness	55.77 ± 10.30	51.93 ± 9.81	6.22	1090	<.001
Conflict	45.03 ± 9.80	44.54 ± 8.92	0.85	1102	.394