

Editorial: Progress and Limitations in the Psychological Study of Craniofacial Anomalies¹

Matthew L. Speltz²

University of Washington

Lynn Richman

University of Iowa

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The term “craniofacial anomalies” (CFAs) refers to a diverse group of congenital disorders including complex syndromes marked by multiple sutural fusions (e.g., Crouzan’s, Treacher Collins, and Aperts syndromes), simple craniosynostoses involving single fusions (e.g., sagittal synostosis), hemifacial microsomia, clefts of the lip and/or palate, and isolated “birth marks” on the face and neck, such as benign vascular nevi (i.e., port-wine stains). Most children with these disorders experience one or more associated complications including feeding and growth difficulties, oral-dental problems, chronic ear infections, speech and language impairments, and multiple structural and cosmetic surgeries for both visible disfigurement and “invisible” dysmorphologies (e.g., cleft palate). Any or all of these factors may produce significant stress and conflict for the child and family.

Various psychological risk factors have been identified that may compromise the child’s psychological adjustment, such as parental guilt or overprotectiveness, stigmatizing social responses to speech impairment or facial disfigurement, and certain neuropsychological limitations (see Barden, 1990; Richman & Eliason, 1993, for reviews of this literature). Although the majority of children with CFA appear to show normal psychosocial development (Speltz, Galbreath,

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²All correspondence should be sent to Matthew L. Speltz, University of Washington, Box 359300-CL-08, Seattle, Washington 98105.

& Greenberg, 1995), the risk of behavior disorders and learning disabilities in this population is higher than expected (Broder & Richman, in press; Speltz, Morton, Goodell, & Clarren, 1993). This suggests that a significant number of these children would benefit from psychological assessment and treatment. In fact, patient care standards set forth by the American Cleft Palate-Craniofacial Association (1993) identified the need for early childhood assessments of developmental status, neuropsychological testing in older children, and various psychological interventions including parent training, presurgery anxiety reduction, and social skills training for children.

Although some CFAs, such as cleft lip and/or palate, occur relatively frequently (e.g., 1 in 1,000 term newborns), many psychologists are unaware of the distinctive psychological sequelae of these disorders and the interventions most likely to reduce the risk of poor outcomes. This may be due, in part, to the infrequent publication of craniofacial research in psychology journals and in part to a history of many anecdotal reports and case histories with minimal empirical data. Existing data have been limited by several methodological problems such as small and diagnostically heterogeneous samples and nonstandardized measures of psychological functioning. In a review and meta-analysis of research on children's physical disorders (Lavigne & Faier-Routman, 1992), only two studies of CFAs met criteria for inclusion.

Another significant problem noted by many researchers in this area has been the absence of studies designed to test a priori hypotheses (Berry, 1995; Eder, 1995). Many early studies of CFAs opportunistically compared children with and without CFA on numerous parent questionnaires and self-reports of psychological adjustment with little theoretical justification. Social-psychological theories of facial attractiveness and attributional processes were first introduced as conceptual frameworks in the late 1970s and 1980s (e.g., Bull & Rumsey, 1988; Tobiasen, 1984). These models led to important advances in the measurement of facial disfigurement (e.g., Tobiasen, 1995) and a better understanding of how dysmorphic appearance may lead to social stigmatization (Langlois, 1995). However, empirical findings have not supported a specific, robust effect of facial dysmorphism on psychological adjustment. Most studies comparing children with *invisible* craniofacial dysmorphologies (e.g., isolated cleft palate) with those having *visible* facial disfigurement (e.g., cleft lip and palate) have found few differences in psychological outcomes (see Speltz et al., 1995, for a review), suggesting that risk factors other than facial disfigurement may be of equal or greater importance (e.g., feeding, speech, or hearing problems).

More recently, developmental models have been proposed to examine the interactions and reciprocal relations among multiple domains of risk and protective factors (e.g., Eder, 1995; Richman & Eliason, 1993; Rubin & Wilkinson, 1995; Speltz, Greenberg, Endriga, & Galbreath, 1994). Most have included child variables (e.g., craniofacial appearance, speech and feeding problems, temp-

erament, cognitive status), parent variables (e.g., personality, well-being, responsiveness), family/social context (e.g., social support, marital functioning), and medical treatment variables (e.g., number, type, and outcome of surgeries). Neuropsychological formulations of CFAs have also shown increased complexity, as indicated by recent work on the differing frequency and course of reading disabilities in children with different cleft diagnoses (Richman & Eliason, 1993) and the cognitive correlates of various craniosynostoses (Kapp-Simon, 1996; Speltz, Endriga, & Mouradian, 1997). There have been improvements in methodology as well. The long-standing emphasis on single variables and cross-sectional designs is being replaced by multimethod assessments (including direct observations of parents and children) and longitudinal and multivariate analyses (e.g., Campis, DeMaso, & Twente, 1995; Hoeksma & Koomen, 1991; Speltz, Endriga, Fisher, & Mason, 1997).

The four articles in this special section of the *Journal of Pediatric Psychology* demonstrate the progress made in recent years, as well as the considerable challenges that remain. Endriga and Speltz (1997) observed the face-to-face, presurgery interactions of mothers and infants with cleft lip and palate, isolated cleft palate, or normal craniofacial morphology. This study is one of the first to examine the behavioral contingencies between mothers and infants with clefts using lag sequential analyses. Endriga and Speltz found that the mothers of infants with cleft palate were more likely than mothers of infants with cleft lip/palate to disengage when the infant was attending to the mother's face. This study is limited by a common problem in the study of clefts: the confounding of gender and diagnosis due to a population gender difference in the incidence of cleft disorders (i.e., more males have cleft lip/palate, more females, cleft palate only) and the difficulty of recruiting subjects of the underrepresented gender.

The study by Pope and Ward (1997) was designed to test theory-driven hypotheses about the correlates of social competence in preadolescent children with CFA, as well as to generate new hypotheses for further research. One of the most interesting findings to emerge from this study—and one with important implications for intervention—was the relation between parental characteristics and preadolescents' social competence: Greater social competence was associated with parents who worried *less* about their child's friendships and actively encouraged their child to interact with peers. This suggests that the social inhibition of some adolescents with CFA may be related, in part, to their parent's anxious communications about the quality of the child's peer relationships. As noted by Pope and Ward, a necessary follow-up to this research is the replication of these findings using a larger sample and multimethod assessments of social functioning.

The Richman and Millard (1997) study represents one of the very few longitudinal studies in this area, and is remarkable in its inclusion of parental

reports of child status for 8 consecutive years. The findings of this study suggest that the behavioral adjustment of children with clefts may vary by age and gender, with females showing increases in both externalizing and internalizing problems with age. An unexpected finding was that measures of three important medical risks associated with clefts (i.e., impaired speech and hearing, and atypical facial appearance) failed to predict behavior problems, after controlling for gender, age, intelligence, and socioeconomic status. This was a very conservative test in that at least one of the covariates—intelligence—has shown strong relations to externalizing problems in samples of children without congenital impairments (e.g., Moffitt & Silva, 1988); and, as the authors note, small samples limited the power of the predictive analyses. Nevertheless, these findings imply that the long-term prediction of psychological outcomes in children with clefts require the measurement of non-cleft-related factors, including parent and family variables.

The final study in this series of papers by Demellweek, Humphris, Hare and Brown (1997) reflects the social-psychological tradition in craniofacial research, with an important methodological advance. Nearly all studies of the attributional responses of children or adults to CFAs have used still photographs of children showing affectively neutral facial poses. However, still photographs cannot provide information about facial movement and communications of affective states, two important sources of information in the perception of facial attractiveness (Berry, 1995; Langlois, 1995). Demellweek et al. assessed the responses of school-age children to “target” children with and without fabricated port-wine stains (PWS) by using videoclips in which target children smiled and briefly talked, turning their head from side to side. The results of their study suggest that peers’ judgments of character, physical attractiveness, and the social impact of PWS are complex and multidimensional, with some effects moderated by gender. Overall, there was little evidence of significant prejudice against children with PWS. The clinical significance of these findings will depend on the extent to which ratings of videoclips can be shown to predict observed social behavior.

In conclusion, the studies in this special section have illustrated some important advances in psychological research on children’s CFAs including micro-analytic coding techniques, hypothesis-driven analyses, prospective longitudinal designs, and the consideration of multiple risk factors. Perhaps the most glaring weakness still evident in this research is one that has limited the study of CFAs for decades: small samples that constrain the power of statistical analyses. Although the CFA samples in these studies are relatively larger than those in many previous studies, the measurement of multiple risk factors has increased the requirements for sample size considerably. Because of the relatively low incidence of these disorders, research informed by multifactorial models requires multicenter investigations, in which the outcomes of children with different combinations of medical and environmental risks can be studied with adequate

statistical power. We hope this special section on craniofacial disorders will help to promote the collaboration necessary to undertake such projects.

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