Social-Emotional Development of Infants and Young Children With Orofacial Clefts

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Children with orofacial clefts are believed to have distinctly elevated risk for a variety of adverse social-emotional outcomes including behavior problems, poor self-concept, and parent-child relationship difficulties. This assumption has been based primarily on theories of facial appearance and social bias, a handful of empirical studies, and clinical impressions. Studies of these children have been limited by methodological problems such as diagnostic heterogeneity, ascertainment bias, and absent or poorly matched control groups. In an attempt to address at least some of these methodological problems, the longitudinal research described in this article examined the developmental course of infants with unilateral cleft lip & palate (CLP) and cleft palate only (CPO). We followed these infants to age 7, with ongoing comparisons to a demographically matched group of typical children. Outcome measures targeted child attachment, maternal/child interaction during feeding and teaching tasks, parent satisfaction with surgical outcomes, parent and teacher behavior rating scales, and child self-concept and behavioral adaptation. Although our findings have provided limited support for the hypothesis that infants and young children with CLP/CPO are at greater risk for social-emotional problems than their peers, we have found that among infants with clefts, early assessment can predict subsequent social-emotional outcomes. In this article, we review theory and data in this area of study, summarize our longitudinal findings, describe our success and failures with respect to methodological rigor, and discuss emerging research and areas for further inquiry. **Key words:** *cleft lip*, *cleft palate*, *craniofacial*, *orofacial cleft*, *social-emotional*

AN orofacial cleft is an opening in the lip or roof of the mouth that results from arrested embryonic development in the first trimester (Siebert, Wiet, & Bumsted, 1998). Clefts are categorized as being "unilateral" or "bilateral" (involving one or both sides of the lip and/or palate) and "incomplete" or "complete" (involving only the soft palate vs both the soft and hard palates, and/or involving only the lip vs the lip and gumline). Orofacial clefts, including cleft lip only (CLO), cleft lip and palate (CLP), and cleft palate-only (CPO), are the second most common birth defect in the United States (Siebert et al., 1998). The incidence of CLO and CLP varies by ethnicity, ranging from 0.3 per 1000 live births among African Americans to 3.6 per 1000 live births in Native Americans (Friedman, Dill, Hayden, & McGillvray, 1992; Lewanda & Jabs, 1994). Males are affected more often than females (eg, CLP 2:1, CLO 1.5:1; Siebert et al., 1998). In contrast, CPO occurs in 1 per 2000 live births with little variation among ethnic groups and increased occurrence among females (4:1; Siebert et al., 1998). Most clefts are "nonsyndromic," meaning that they occur in the absence of other malformations and/or significant impairment of the central nervous system (Johnston & Bronsky, 1995; Jones, 1988). Diagnostic heterogeneity is common in psychologic research on craniofacial anomalies (CFA) such that children with nonsyndromic clefts are often mixed with children having other types of

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CFA (eg, craniosynostosis, hemifacial microsomia), including some with a known genetic syndrome (eg, Aperts).

Although there are no widely accepted models of inheritance, it is believed that genetic processes play a role in nonsyndromic clefting. Prenatal environmental factors, such as maternal smoking, alcohol use, exposure to anticonvulsant medications, and maternal nutritional status, have also been implicated (Prescott & Malcolm, 2002). Contemporary research has examined the interplay between genetic vulnerability (eg, "susceptibility genes") and these environmental precipitants (Wyszynski, Beaty, & Maestri, 1996).

Orofacial clefts can be identified via ultrasound in utero. However, many cases are missed using this technology, and often the diagnosis is not made until delivery (Johnson & Sandy, 2003). As with any congenital deformity, learning that one's child has a cleft can be devastating for parents. Parents' immediate reaction to diagnosis has not been well studied; however, clinical impressions and the results of existing descriptive studies suggest that parents have vivid recollections of receiving the diagnosis and the sensitivity with which it was handled by medical professionals (eg, Strauss, Sharp, Lorch, & Kachalia, 1995).

Following delivery, families are generally referred to a specialty craniofacial clinic for management of their child's care. Immediate concerns typically focus on feeding and physical growth, as children with clefts have difficulty "latching" and developing the suction necessary for breastfeeding, thus requiring accommodating devices (eg, specially designed nipples for bottles, syringe with rubber tubing, etc). With development, children undergo a series of surgical procedures to repair their cleft lip and/or palate. Surgical repair of the cleft lip is typically performed when the infant is 2 to 5 months of age, and cleft palate repair is usually performed when children are between 12 and 18 months of age. Subsequent procedures and surgical revisions are scheduled according to patient needs, including developmental changes in facial structure and appearance. Complications such as repeated ear infections, dental/orthodontic concerns, airway obstruction, and difficulty with speech-language development are common. Thus, the craniofacial team monitors these children on a regular basis to address concerns as they arise.

In clinical and case study literature dating back to the 1950s, children with clefts have been noted to show elevated rates of problems in social-emotional development. The highly visible and socially stigmatizing nature of the conditions, the anxiety and distress often reported by these children's parents, and the sequelae and stressful demands of medical treatment have all been cited as factors contributing to such problems. However, research findings have been mixed and, in some cases, counterintuitive.

In the first part of this article, we discuss this research, focusing on the period from birth to early elementary school. Using electronic databases (ie, Psychinfo, Medline) and a review of journals that publish studies of this population (eg, *The Cleft Palate-Craniofacial Journal*), we identified studies that (1) included primarily young children with orofacial clefts and their families and (2) focused on early childhood development, parent-child relationships, and child social and emotional adjustment. In a few cases, studies of heterogeneous CFA samples were included for review if they included an adequate proportion of children with clefts, and represented a unique contribution to the literature. These findings are described and the methodological limitations that influence their interpretation discussed. In the second part of the article, the findings from a longitudinal, case-controlled study recently completed by our research team are summarized. This study followed the development of children with and without orofacial clefts from early infancy through age 7. We discuss our successes and failures in addressing the methodological limitations of earlier studies. We conclude with a discussion of the clinical implications of our findings and those of other

researchers, as well as directions for future research.

SOCIAL-EMOTIONAL DEVELOPMENT—REVIEW OF LITERATURE

Parent coping and parent-child relationships

For parents, having a child with an orofacial cleft raises anxieties about the cause of the condition (eg, *did I do something to cause this?*), concern about the reactions of peers and others to their child (eg, *will he be teased?*), and questions regarding the developmental implications of the disorder (eg, *will my child be mentally retarded?*) (Dar, Winter, & Tal, 1974; Drotar, Baskiewicz, Irvin, Kennell, & Klaus, 1975). There is emerging research on parents' response to the "diagnostic event." That is, the point at which parents first hear about and begin to cope with their child's cleft diagnosis. There tends to be a sharp contrast between the information that parents recall being given during the diagnostic visit and the information that they desired (Byrnes, Berk, Cooper, & Marazita, 2003; Strauss et al., 1995; Young, O'Riordan, Goldstein, & Robin, 2001). In particular, Young et al. (2001) found that parents expressed a desire for more information about feeding, other factors that may be associated with clefting (eg, developmental delay), and normal, as well as abnormal, findings from their child's examination (eg, "her muscle tone is normal"). Parents also wanted healthcare providers to use accurate and sensitive descriptions of their child's deformity (eg, "cleft lip" vs "birth defect" or "harelip") and to provide reassurance that their child's cleft was not their fault and that their child was not in pain.

As might be expected, parents' concerns evolve over time. Initial worries focus on the pragmatics of feeding, timing of lip repair surgery, and possible embarrassment upon introducing the newborn child to others. Later, concerns turn to determining the cause of the deformity, the duration of craniofacial care, and the risk for future pregnancies (Dar et al., 1974). A subset of parents report experiencing depressive symptoms, increased stress, lower evaluations of competence, and increased marital conflict related to their child's cleft diagnosis (Dolger-Hafner, Bartsch, Trimbach, Zobel, & Witt, 1997; Speltz, Armsden, & Clarren, 1990). However, increased parental distress is not consistently found relative to the parents of healthy controls (Campis, DeMaso, & Twente, 1995; Krueckeberg & Kapp-Simon, 1993; Pelchat, Bisson, Ricard, Perreault, & Bouchard, 1999), and the parents of children with CFAs may report less distress than the parents of children with other medical conditions (eg, Down syndrome, congenital heart disease; Pelchat et al., 1999). Recently, Pelchat et al. (1999) have published promising data from a study investigating a psychosocial intervention intended to facilitate parents' adaptation to their child's cleft condition, with results showing the benefits of intervention both immediately and at follow-up.

Given the challenges that a cleft condition might pose to parent and child adaptation, several groups have studied parent-child relationships in cleft populations, with an emphasis on parents' sensitivity, responsiveness, and engagement. This work has also investigated objective ratings of children's "cues" to parents during interaction, to determine whether children with clefts may provide fewer, or less clear, cues in the regulation of their caregivers' attention. Studies have shown that infants with orofacial clefts and their parents are less active, playful, and sensitive during parent-child interactions than controls (Barden, Ford, Jensen, Rogers-Salyer, & Salyer, 1989; Field & Vega-Lahr, 1984; Koomen & Hoeksema, 1992; Wasserman & Allen, 1985; Wasserman, Allen, & Solomon, 1986). For example, Barden et al. (1989) found that infants with heterogeneous CFAs spent less time than controls looking, touching, and otherwise engaging their mothers during interaction. The

infants with CFAs also cried more and showed more gaze aversion (ie, turning away in response to mothers' gaze). These infants' mothers spent less time in the en face position, provided less physical contact, smiled/laughed less, and spent less time demonstrating a toy.

Studies of toddler/preschooler-parent dyads yield mixed results. Research by Wasserman and colleagues (eg, Allen, Wasserman, & Seidman, 1990; Wasserman & Allen, 1985; Wasserman et al., 1986) indicated that mothers *increased* their level of activity and engagement in interactions with their toddlers/preschoolers with clefts, perhaps in response to perceived and/or actual delays in their child's development. These parents were more active "teachers," using a high rate of behavioral commands, requests, and distractions (Allen et al., 1990), and tended to rely on negative and ineffective limit setting strategies (Wasserman et al., 1986). The overall impression is of a controlling and overly protective parent, seeking to elicit performance from his or her child. However, other research groups have found few, if any, differences between parent-child dyads with and without clefts in free play or teaching interactions (Chapman & Hardin, 1991; Pelchat et al., 1999; Speltz, Armsden, & Clarren, 1990) or in the self-reported parenting styles of parents with and without clefts (Krueckeberg & Kapp-Simon, 1993). It is important to note that these research groups have each used slightly different methods, and the discrepant findings might reflect differences in observational tasks and coding systems. In studies using the well-known Strange Situation procedure (Ainsworth, Blehar, Waters, & Wall, 1978), few, if any, differences in attachment status have been found in infants with clefts relative to healthy controls (Koomen & Hoeksema, 1993; Wasserman, Lennon, Allen, & Shilansky, 1987). Indeed, these children may be more likely to have a secure attachment than other groups with chronic medical illnesses (eg, those with neurological impairments; Clements & Barnett, 2002) (Table 1).

Infant and early child development

Several studies show that infants and toddlers with CLP and CPO score within the low average to average range on clinician-administered measures of mental and motor development (eg, the Bayley Scales of Infant Development [BSID]; Jocelyn, Penko, & Rode, 1996; Kapp-Simon & Krueckeberg, 2000; Starr, Chinsky, Canter, & Meier, 1977) and parent report scales (eg, Neiman & Savage, 1997). A higher percentage than expected score within the "developmentally delayed" range (ie, roughly 15% vs 2% in the normative population; Kapp-Simon & Krueckeberg, 1996). Comparisons among cleft groups suggest that those with CPO receive the lowest scores, followed by children with CLP and CLO, who tend to receive roughly average scores (Kapp-Simon & Krueckeberg, 1996; Starr et al., 1977). Young children with physical malformations in addition to clefts may be at particular risk (Swanenburg De Veye, Beemer, Mellenbergh, Wolters, & Heineman-De Boer, 2003). In one of the few studies including a demographically matched control sample, Jocelyn et al. (1996) found that children with CLP received significantly lower mental and motor development BSID scores than controls, though still well within the average range of BSID test norms (Table 2).

Child emotional and behavioral adjustment

Research on the emotional and behavioral adjustment of children with clefts has produced conflicting findings. The most commonly reported finding, primarily from the work of Richman and colleagues (Richman, 1976, 1978, 1983; Richman & Harper, 1979; Richman & Millard, 1997), is that children with clefts are more socially withdrawn and inhibited than controls, particularly in the classroom. This appears to be especially true for children with CPO (vs unilateral or bilateral CLP; Richman & Millard, 1997). Other studies have failed to find statistically significant case/control differences in

Table 1. Studies of parent coping and parent-child relationships*

Authors	Sample	Domains Assessed and Research Design	Results
Tisza and Gumpertz (1962)	Unspecified sample of mothers whose children had CLP/CPO	Qualitative description of maternal responses following birth of a child with a congenital deformity	Idiosyncratic beliefs regarding origins or reasons for child's deformity, description of bereavement/ grieving process following child's birth
Dar et al. (1974)	Prospective study of parents (numbers of mothers and fathers unspecified) of children w/CLP ($n = 21$) and CLO ($n = 3$). Retrospective study of parents of children w/CPO ($n = 6$), CLP ($n = 13$), and CLO ($n = 8$)	Parents' reactions and attributions immediately following the birth of a child w/a cleft and again 12 mos later (prospective study), and parents' recollections when their child was age 1–6 years (retrospective study)	Immediately after birth, parents were most concerned about the home care their child would require (eg, feeding modifications), when their child would require surgery, embarrassment upon introducing their child to others, and possible effects on cognitive function. At follow-up and in the retrospective study, parents were more concerned w/the duration of their child's treatment, the cause of their child's malformation, and risks for future pregnancies. When asked what they attributed the child's malformation to, parents reported a variety of idiosyncratic beliefs (eg, medicines used during pregnancy, looking at a man w/a cleft, divine influence, and parental sin) *(continues)*

Table 1. Studies of parent coping and parent-child relationships*(*Continued*)

Authors	Sample	Domains Assessed and Research Design	Results
Speltz et al. (1990, 1993)	Mothers of toddlers w/heterogeneous CFAs ($N = 33$; CPO $n = 12$, CLP $n = 11$, sagittal craniosynostosis $n = 10$) and controls ($n = 22$). At follow-up roughly 4-years later, sample included $N = 23$ mothers of cases (CPO $n = 7$, CLP $n = 9$, sagittal craniosynostosis $n = 7$) and 10 controls	Mothers' self-reported parenting stress, emotional well-being, marital satisfaction, social support, and developmental expectations	Compared w/controls, mothers of children w/CFAs reported higher parenting stress, lower self-competence, and higher marital conflict. At follow-up, the mothers of children w/CFAs reported poorer emotional health and, for mothers of infants w/CLP, less satisfaction w/their social support
Krueckeberg and Kapp-Simon (1993)	Parents (numbers of mothers vs father unspecified) of children w/heterogeneous CFAs ($N = 30$; CPO $n = 8$, CLO $n = 7$, CLP = 5) and healthy controls ($N = 22$)	Parents' self-reported parenting stress, parenting style, and social support; children's social skills	Parents of children w/CFAs did not differ from controls in parenting stress, parenting style, or social support. Parents whose children had "visible defects" reported greater social support and reported a more nurturing and less restrictive parenting style. For all groups, parenting stress predicted children's social skills

(continues)

Table 1. Studies of parent coping and parent-child relationships*(*Continued*)

Authors	Sample	Domains Assessed and Research Design	Results
Campis et al. (1995)	Mothers of children w/heterogeneous CFAs ($N = 77$; CLP, CPO, CLO $n = 33$, facial microsomia $n = 18$, unilateral coronal craniosynostosis $n = 11$, vascular anomalies $n = 6$, syndromic craniosynostosis $n = 4$, frontonasal dysplasia $n = 4$, and acquired facial deformity $n = 1$)	Mothers' psychologic functioning (depression, anxiety, parenting stress, and social support)	Mothers of children w/CFAs scored w/in the average range on all measures
Strauss et al. (1995)	Parents (90 mothers, 10 fathers) of children w/CLO ($n = 4$), CLP ($n = 46$), and CPO ($n = 50$)	Maternal perceptions of diagnostic process	Parents reported desire for more information regarding possible developmental delay, for more opportunity to talk and share their feelings with care providers, for their physician to show more caring and confidence, and referral to other parents of cleft children
Dolger-Hafner et al. (1997)	Parents (41 mothers, 39 fathers) of children with CLO/CLP ($N = 55$)	Parental reactions to the birth of a child with an orofacial cleft	Most parents had not anticipated possibility of child deformity; frequent reports of guilt, depression, and social avoidance
Pelchat et al. (1999)	Parents of infants w/CLO/CLP ($n = 38$), Down	Parents' self-reported parenting stress, stress appraisal, and	Parents of infants w/Down syndrome and congenital heart (*continues*)

Table 1. Studies of parent coping and parent-child relationships*(*Continued*)

Authors	Sample	Domains Assessed and Research Design	Results
	syndrome ($n = $ 32), congenital heart disease ($n = 36$), or no disability ($n = 38$)	psychologic distress (in conjunction w/intervention-outcome study)	disease reported greater stress than CLO/CLP and non-disabled controls. For all groups, mothers reported greater stress and psychologic distress than fathers
Slade, Emerson, and Freedlander (1999)	Mothers of infants with CLP/CLO ($N = 33$)	Maternal psychopathology, feelings of attachment, and parental competence as a function of timing of cleft repair (before/after 3 mos)	No differences as a function of timing of surgery. For both groups, symptoms decreased over time.
Pelchat et al. (1999)	Parents of children with CLP ($n = $ 43) or Down syndrome ($n = $ 31); assigned to either treatment or non-treatment control groups	Efficacy of an intervention program intended to improve parents' adaptation to their child's condition	Parents who received intervention showed better adaptation immediately following treatment and at follow-up assessment when their child was 12 and 18-months old
Young et al. (2001)	Parents (36 mothers, 4 fathers) of children w/CLP ($N = 40$)	Parents' recollection of diagnostic visit and preferences for diagnostic feedback	Parents reported a desire for more information about: feeding, feedback about normal and positive findings from their child's exam, and possibility of associated malformations/developmental delay. Parents also reported a desire for clinicians to use proper terminology, to provide reassurance that child was not in pain, and to reassure them that the child's cleft was not their fault.

Table 1. Studies of parent coping and parent-child relationships*(*Continued*)

Authors	Sample	Domains Assessed and Research Design	Results
Byrnes et al. (2003)	Parents (97 mothers, 1 father) of children w/CLP/CLO ($n = 60$) or CPO ($n = 38$)	Maternal perceptions of diagnostic process (same measure as Strauss et al., 1995)	Parents expressed dissatisfaction with several elements of diagnostic feedback provided. Desire for more information regarding developmental implications, opportunity to meet with other parents of cleft children, more comfort from child's physician
Field and Vega-Lahr (1984)	Mothers of children w/CLP ($n = 12$) vs control ($n = 12$)	Infant cues and maternal sensitivity	Fewer clear infant cues in CLP group, mothers of CLP infants less engaged/responsive than controls
Wasserman et al. (1986)	Mothers of children w/heterogeneous CFAs or orthopedic impairment (CPO $n = 3$; CLP $n = 3$; CLO $n = 1$; other $n = 11$) vs premature ($n = 15$) vs controls ($n = 18$)	Mothers' limit-setting	Mothers of CFA/orthopedically impaired children used more negative/ less effective limit setting strategies than mothers of premature children and mothers of controls.
Wasserman et al. (1987)	Children w/heterogeneous CFAs or orthopedic impairment ($n = 36$) vs controls ($n = 46$)	Infant attachment security assessed using Strange situation	No differences between CFA/orthopedic and control
Barden et al. (1989)	Children w/heterogeneous CFAs ($n = 5$) vs control ($n = 5$)	Maternal self-reported satisfaction; maternal nurturance during mother-child interaction	Mothers of infants with CFA reported higher parenting satisfaction and life satisfaction than controls; Mothers of infants with CFA showed less nurturing behavior during interactions than controls

Table 1. Studies of parent coping and parent-child relationships*(*Continued*)

Authors	Sample	Domains Assessed and Research Design	Results
Speltz et al. (1990)	Mothers of children w/heterogeneous CFAs ($N = 33$; CPO $n = 12$, CLP $n = 11$, sagittal craniosynostosis $n = 10$) vs control ($N = 10$)	Maternal responsiveness during free play and teaching task interactions; toddler responsiveness during interaction	No differences between CFA and control groups
Allen et al. (1990)	Children w/ heterogeneous CFAs ($n = 37$; CLP $n = 10$, CLO $n = 1$, CPO $n = 4$) vs demographically matched control ($n = 44$)	Observation of maternal responsiveness, teaching, control, and encouragement; Child compliance, initiative, distractibility	Children with CFAs showed less social initiative than controls and were more cooperative during a clean-up activity. During teaching interaction, the mothers of children w/CFAs were more active and controlling than control mothers
Chapman and Hardin (1991)	Mothers of children w/CLP (n = 13) vs control (n = 13)	Maternal language input	Few differences between CLP and control
Koomen and Hoeksema (1992)	Mothers of children w/CLP (n = 33) vs matched control (n = 34)	Maternal sensitivity during play interaction	Mothers of CLP infants less sensitive than controls
Koomen and Hoeksema (1993)	Mothers of children w/CLP hospitalized at 9.5 mos ($n = 14$) vs 12.5 mos ($n = 13$), vs control ($n = 14$)	Infant attachment security and attachment behaviors using Strange situation	No differences in attachment security, CLP infants showed greater avoidance
Clements & Barnett (2002)	Children w/CFAs or limb deformity (CLO $n = 14$, CLP $n = 10$, CPO $n2$, Other CFA $n = 6$, limb deformity1) vs neurologic impairment ($n = 39$; eg, cerebral palsy)	Infant attachment security using Strange situation and Attachment Q-Sort, and parenting quality	Higher parenting quality ratings were associated w/visible malformations. Children w/CFAs and those w/visible malformations were more likely to be securely attached.

*CLP indicates cleft lip and palate, CPO, cleft palate-only; and CFA, craniofacial anomalies.

Table 2. Studies of infant/early childhood development*

Authors	Sample	Domains Assessed and research design	Summary of results
Starr et al. (1977)	Children w/CPO (*n* = 24), CLP (*n* = 31), and CLO (*n* = 20). A sub-sample of 28 children were tested at all time points: CPO (*n* = 8), CLP (*n* = 12), and CLO (*n* = 8)	Clinician assessed mental and motor development (BSID) in a mixed cross-sectional and longitudinal design	No group differences in mental or motor development at 6 and 12 mos, all groups scored in an average range compared to BSID norms. At 18 mos, CPO and CLP scored in low average range in mental and motor development and significantly lower than CLO. At 24 mos, CLP scored in low average range in mental and motor development and significantly below CPO and CLO. Longitudinal analyses did not reveal w/in group differences by age.
Jocelyn et al. (1996)	Children w/CLP (*n* = 16) & demographically matched controls (*n* = 16).	Clinician assessed mental and motor development (BSID), and expressive/ receptive language in longitudinal study	Children w/CLP scored significantly below controls in mental and motor development at 12 and 24 months, with scores ranging from low average to average compared to test norms. Children w/CLP also scored significantly lower than controls on measures of expressive/receptive language
Neiman and Savage (1997)	Children w/CLO (*n* = 48), CPO (*n* = 46), and CLP (*n* = 92).	Parent-reports of infant development (KID, MCDI) in longitudinal study	At 5 and 13 mos, no significant group differences, though children were more likely than normative group to score in "at-risk" range. At 25 and 36 ms, all groups scored roughly w/in average range w/a few exceptions (eg, CPO children more likely to score "at-risk in expressive language

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Table 2. Studies of infant/early childhood development*(*Continued*)

Authors	Sample	Domains Assessed and research design	Summary of results
Kapp-Simon and Krueckeberg (2000)	Children w/CLO ($n = 19$), CPO ($n = 59$), CLP ($n = 88$), and Pierre Robin Syndrome ($n = 14$). A sub-sample of 85 children (CLO $n = 6$, CPO $n = 28$, CLP $n = 45$, Pierre Robin Syndrome $n = 6$) were followed longitudinally	Clinician assessed mental development (BSID) in a mixed cross-sectional and longitudinal design	All groups scored in the average range compared to BSID norms. The 18 and 24-month groups scored significantly lower than 6 and 12-month groups. Children w/CLO scored significantly higher than those w/CLP and Pierre Robin sequence. In longitudinal analyses, scores declined significantly over time. At 24 months, 15.3% of the sample scored in the "developmentally delayed" range compared w/test norms
Swanenburg De Veye et al. (2003)	Children w/CLP ($n = 32$) and CLP + malformation ($n = 30$); CLO ($n = 47$) and CLO + malformation ($n = 9$); & CPO ($n = 16$) and CPO + malformation ($n = 16$)	Clinician assessed mental and motor development (BSID, Dutch version)	Children w/CLO scored significantly higher than those w/CLP and CPO in mental development, though all groups w/in average range compared to test norms. No group differences in motor development. Children w/malformations scored significantly lower in mental and motor development than those w/out malformations. Scores for children w/"minor malformations" w/suspected developmental implications those w/"major malformations" scored in low average to mildly delayed range compared w/test norms

*CPO indicates cleft palate-only; CLP cleft lip and palate; and CLO, cleft lip only.

behavioral outcomes, although a sizable minority of those with clefts had clinically significant behavior problems (eg, Speltz, Morton, Goodell, & Clarren, 1993). There is evidence suggesting that children with clefts who are referred for mental health evaluations of behavioral problems are frequently misdiagnosed as having attention-deficit/hyperactivity disorder (ADHD), even when their symptoms are better accounted for by another disruptive behavior disorder, learning disability, or anxiety (Richman, Ryan, Wilgenbusch, & Millard, 2004).

Behavioral and emotional adjustment for children with clefts may vary as a function of age and gender. In a cross-sectional sample of preschool, elementary school, and junior high school students with CLP, Schneiderman and Auer (1984) found that mothers, fathers, and teachers reported more behavior problems among older age groups. Longitudinal studies suggest increasing internalizing and externalizing behaviors in middle childhood (ie, ages 7–12 years) especially among girls with clefts (Richman & Millard, 1997; Speltz, Morton, Goodell, & Clarren, 1993). Researchers investigating predictors of outcome have found that speech problems predict internalizing symptoms for those with CPO while facial disfigurement predicts emotional/behavioral outcomes for those with CLP. Maternal functioning (eg, parenting stress, psychologic functioning) has proven to be a robust predictor of outcome that may be superior to the child's objective medical status (eg, Campis et al., 1995; Speltz et al., 1993). This highlights the importance of better understanding parents' adjustment to their child's diagnosis (Table 3).

Self-concept

Self-concept has been a primary interest for researchers investigating social-emotional outcomes in children with clefts. The underlying hypothesis has been that these children internalize the differential treatment they receive from others (presumably, negative responses to physical appearance or speech) and therefore develop negative self-perceptions. Commonly assessed domains of self-perception include appearance, friendships, and problem-solving ability and intellectual competence. A few studies have shown that young children with clefts scored lower on measures of self-concept than typically developing peers (Broder, Smith, & Strauss, 1994; Broder & Strauss, 1989; Kapp-Simon, 1986). Children with "visible" clefts (CLO, CLP) tend to report less satisfaction with their appearance than those with "invisible" clefts of the palate only (Broder et al., 1994; Broder & Strauss, 1989).

However, such differences are not consistently found, and do not appear to generalize to all areas of self-perception. For example, Leonard, Brust, Abrahams, and Sielaff (1991) found that self-concept scores for youths with CLO, CLP, and CPO were in the moderate to high range relative to test norms and none of their participants reported abnormally low self-concept. In heterogeneous CFA samples, Krueckeberg, Kapp-Simon, and Ribordy (1993) and Speltz et al. (1993) found that children with CFA generally scored within the average range on a self-concept measure and differed little from control children. In fact, Krueckeberg et al. (1993) found that girls with CFA had above-average self-concept compared with test norms, and received higher scores than did girls in the control sample. In the study cited above by Broder et al. (1994), although children with clefts reported concerns regarding their appearance, they also reported having more friends than typically developing children, and expressed greater interest in playing with peers rather than playing alone (Table 4).

Peer relations and social perception

Social-psychologic studies of the relation between physical attractiveness and social perception and acceptance (see Langlois et al., 2000, for a review) have influenced the work of several investigators interested in the peer relationships of children with orofacial

Table 3. Studies of child emotional and behavioral adjustment*

Authors	Sample	Domains assessed and research design	Results
Richman (1976)	Children w/CPO (*n* = 29), CLP (*n* = 15), and healthy controls (*n* = 44)	Teacher-reported behavior problems (Behavior Problem Checklist)	Children w/clefts had higher internalizing behavior scores than controls. No difference in teacher-reported conduct problems
Richman (1978)	Children w/CPO (*n* = 72) and CLP (*n* = 64)	Parent and teacher reported behavior problems (Behavior Problem Checklist)	Teachers rated boys w/clefts as having fewer conduct problems and more internalizing behaviors than parents. Teachers rated girls w/clefts as showing more internalizing symptoms than parents
Richman and Harper (1979)	Children w/CLP (*n* = 45) and orthopedic impairment (eg, cerebral palsy, meningomyelo-cele; *n* = 45)	Self-report of personality (Missouri Children's Picture Series)	Boys w/CLP scored higher than those w/orthopedic impairment on "maturity" and "inhibition" subscales, and lower on "aggression," "activity level," and "somatization" subscales. Girls w/CLP also scored higher than those w/orthopedic impairments on "maturity" and "inhibition" scales, and lower on "masculinity/femininity."
Schneiderman and Auer (1984)	Children w/CLP (*n* = 58)	Parent and teacher rated behavior problems (Behavior Problem Checklist) in a cross-sectional study	Behavior problems (conduct problems, internalizing problems, socialized delinquency) increased w/age between preschool and junior high. Boys showed more conduct problems than girls
Speltz et al. (1993)	Children w/heterogeneous CFAs (*N* = 23; CPO *n* = 7, CLP *n* = 9, sagittal craniosynosto-sis *n* = 7) and controls (*N* = 10)	Parent and teacher reported behavior problems (Child Behavior Checklist and Teacher Report Form) in a follow-up study	Girls w/CFAs received higher Total CBCL scores than boys w/CFAs and control girls; girls w/CFAs also received higher scores than control girls in externalizing and internalizing domains. 18% of children w/CFAs had scores at or above the clinical cut-off on both the CBCL & TRF. Observed mother-child interactions predicted children's CBCL scores.

(*continues*)

Table 3. Studies of child emotional and behavioral adjustment*(*Continued*)

Authors	Sample	Domains assessed and research design	Results
Richman and Millard (1997)	Children w/CLO ($n = 21$) & CLP ($n = 23$)	Parent-reported behavior problems (Behavior Problem Checklist) in a longitudinal study	For girls, internalizing and conduct problem behaviors increased between ages 4 and 12. Among boys, conduct problems decreased with age. Boys and girls with clefts received higher scores than a normative sample. Internalizing behaviors for both genders were predicted by speech defectiveness ratings.
Millard and Richman (2001)	Children w/unilateral CLP ($n = 25$), bilateral CLP ($n = 21$), or CPO ($n = 19$)	Parent teacher rated behavior problems (Pediatric Behavior Scale); child self-report of anxiety and depression (Revised Children's Manifest Anxiety Scale, Reynolds Child and Adolescent Depression Scale); child self-perception (interview); & facial/speech ratings	Parents- and teachers-rated children w/CPO as more depressed and anxious than other groups; also rated as having more learning problems. Children with unilateral CLP-rated themselves as less depressed than others. For Unilateral and bilateral CLP groups, self-reported depression, anxiety, and self-perception positively related to facial attractiveness ratings, not speech ratings. For CPO, self-reported depression, anxiety, and self-perception related to speech ratings, not facial attractiveness ratings
Richman et al. (2004)	Children w/CLP ($n = 177$); diagnostic evaluation & follow-up w/a sub-sample ($n = 32$) who had been diagnosed w/ADHD	Parent- and teacher-reported behavior problems (Pediatric Behavior Scale); language, memory, visual-spatial, and visual-motor skills; attention and impulsivity during a continuous performance task	18% of the sample had been diagnosed w/ADHD and were receiving stimulant medication. Using research criteria (consistent with *DSM-IV*), 69% did not meet diagnostic criteria for ADHD. Many had undiagnosed symptoms of a learning disorder (66%), an anxiety disorder (38%), oppositional defiant disorder (16%), and/or conduct disorder (6%)

*CPO indicates cleft palate-only; CLP, cleft lip and palate; CLO, cleft lip only; ADHD, attention-deficit/hyperactivity disorder; and *DSM-IV*, *Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition*.

Table 4. Studies of child self-concept*

Authors	Sample	Domains Assessed	Results
Kapp-Simon (1986)	Children w/CLO ($n = 4$), CLP ($n = 31$), CPO ($n = 15$), and healthy controls ($n = 172$)	Multidimensional self-concept (total score, personal, social, intellectual)	Children with clefts more likely to report low self-concept than controls on total, personal, and social domains; no differences between cleft groups
Broder and Strauss (1989)	Children w/CLO ($n = 13$), CLP ($n = 13$), CPO ($n = 14$), and healthy controls ($n = 18$)	Multidimensional self-concept (total score, personal, social, intellectual)	Children w/CLO and CLP had lowest personal, social, and total self-concept scores followed by those w/CPO and controls. Children w/clefts more likely than controls to report "low" self-concept in personal and social domains relative to test norms
Broder et al. (1994)	Children w/CLO/CLP ($n = 272$), CPO ($n = 159$), & and healthy controls ($n = 128$)	Multidimensional self-concept (appearance, problem solving ability, peer relations)	Boys w/"visible" defects (CLO/CLP) reported lower appearance ratings than boys w/"invisible" defects and controls. Young girls (ages 5–9) w/CLO/CLP reported lower appearance ratings than girls w/CPO and controls; no differences emerged for preadolescent-adolescent girls (ages 10–18). Children w/clefts reported having more friends than controls Children w/CPO reported lower ability to solve difficult problems than those w/CLO/CLP & controls
Krueckeberg et al. (1993)	Children w/ heterogeneous CFAs ($N = 30$; CLO $n = 7$, CLP $n = 5$, CPO $n = 8$) and healthy controls ($n = 22$)	Self perception (physical, cognitive, peer acceptance, and maternal acceptance)	No differences between CFA and control groups overall. Girls w/CFAs showed better self perception than controls in maternal & peer acceptance; & better self perception than males w/CFAs in physical acceptance. No differences observed for "visible" vs "invisible" defects

Table 4. Studies of child self-concept*(*Continued*)

Authors	Sample	Domains Assessed	Results
Speltz et al. (1993)	Children w/ heterogeneous CFAs ($N = 23$; CPO $n = 7$, CLP $n = 9$, sagittal craniosynostosis $n = 7$) and controls ($N = 10$)	Global self-concept	No differences between CFA and control groups. Self-concept was predicted by maternal teaching behavior during observed parent-child interactions
Leonard et al. (1991)	Children and adolescents w/CLP ($N = 105$)	Multidimensional self-concept (behavior, intellectual and school status, physical appearance and personal attributes, anxiety, popularity, and happiness and satisfaction	Majority of children had average-high self-concept scores relative to test norms. Adolescent girls had lower self-concept than other groups (young girls, young boys, adolescent boys). All groups had lower scores on "physical appearance" items vs "personal attributes" items

*CLO indicates cleft lip only; CLP, cleft lip and palate; CPO, cleft palate-only; and CFA, craniofacial anomalies.

clefts. This research has emphasized the social skills of these children (eg, Krueckeberg et al., 1993), as well as the influence that clefts have on the social perceptions held by nonaffected individuals (eg, Schneiderman & Harding, 1984). Social skills measures have revealed subtle but important differences between young children with and without clefts. Krueckeberg et al. (1993) interviewed children about hypothetical social encounters and found that those with CFAs gave fewer "friendly" responses than did controls. In a recent observational study, Slifer et al. (2004) found that children with clefts were less likely than controls to state a choice/preference during social interaction and to answer a peer's questions. The authors suggest that these features may reflect a passive and self-conscious pattern of interaction. In a study by Chapman, Graham, Gooch, and Visconti (1998), preschool and early school-aged children with CLP did not differ significantly from controls in overall level of conversational skill.

However, a substantial number of these children (50% of preschoolers, 20% school-aged children) showed a less assertive style of communication than controls.

Studies of social perception and acceptance have provided more consistent results, with several authors finding that children and adults rate youths with CFAs as less attractive than controls. Consistent with the broader literature on attractiveness, lay observers also tend to provide negative evaluations of these children's personality and other attributes. Krueckeberg et al. (1993) found that adults rated children with CFAs as less attractive than controls. Child raters in the study by Schneiderman and Harding (1984) were more likely to choose adjectives such as dirty, mean, weak, frightened, slow, and sad for children with clefts versus controls, with those having bilateral CLP receiving the most negative evaluations. Similarly, Tobiason (1987) found that children aged 8 to 16 rated described youths with CLP as less friendly, unpopular,

less intelligent, unattractive, and less likely to be a chosen friend than noncleft children. Ratings did not differ by age or gender of the rater.

Speech quality (eg, hypernasality) is also of concern, and has been hypothesized to influence listeners' perception of other child attributes (eg, personality, intellectual competence). However, in a study by Berry, Witt, March, Pilgram, and Eder (1997), child raters of speech samples were not overly sensitive or critical of the speech of children with CLP or CPO with repaired clefts (Berry, Witt, Marsh, Pilgram, & Eder, 1997). Perhaps not surprisingly, a group of speech-language pathologists rating the same speech samples detected subtle speech differences and often endorsed an item suggesting that the children required further evaluation and/or treatment (Table 5).

Summary and critique

There is now a sizable body of research examining social-emotional development in young children with clefts and their parents. The core assumption underlying this research is that these children have elevated risk for poor developmental and social-emotional outcomes. In some cases, research has shown this to be an accurate characterization. However, findings have been quite variable and, when differences have emerged, they often reveal small effects of questionable clinical or ecological significance.

The methodology used in this work may contribute to these variable findings, possibly minimizing group differences. In most studies, sample sizes have been small, limiting statistical power and making it difficult to examine sociodemographic variables (eg, socioeconomic status [SES]). This is a significant limitation, in light of work showing that risk factors for developmental and social-emotional outcomes tend to be cumulative and that children's quality of care is related to social risk (Sameroff, Gutman, & Peck, 2003). A related problem in this research is the ascertainment of cases, which can introduce sample bias for very different reasons. For ex-

ample, children and families at the highest social risk might be the least likely to take part in research, because of family instability and poor access to and follow-up with medical providers. Alternatively, it may be that families who maintain close contact with a major medical center (and are therefore more available for research participation) have children with more severe cleft conditions or associated problems than nonparticipating families of children with clefts. Both possibilities could restrict the range of participants seen and limit power to detect group differences. Early studies in this area tended to recruit diagnostically heterogeneous samples, often including youths with CFAs other than nonsyndromic orofacial clefts (eg, craniosynostosis, hemifacial microsomia). Given that these conditions can have different manifestations and medical and developmental implications, this strategy could "wash out" effects for specific subgroups.

It is also notable that most studies in this area have been cross-sectional, featuring single assessments of children with clefts, therefore limiting our understanding of the developmental course of this disorder. Few published studies have included multiple measures of the constructs studied (eg, self-concept, parent-child relationship) and most have relied exclusively on parent or self-reports of constructs rather than including observational measures. Although self-rating or parent rating scales and checklists can provide very efficient and psychometrically sound assessment, they are also prone to social desirability and/or "halo" effects (Martin, 1988; Martin, Hooper, & Snow, 1986). For example, children and youth with clefts may underreport difficulties with peers, or overrate their friendships or self-image because they are embarrassed to do otherwise. Caregivers may seek to "protect" their child by underreporting behavior problems observed in the home. Observations alone can be equally problematic, as it is sometimes impossible to keep observers "blind" to children's case status and this may lead to biased ratings. The combination of both methods is ideal.

Table 5. Studies of peer relations and social perception*

Authors	Sample	Domains assessed	Results
Schneiderman and Harding (1984)	78 child raters ages 7–10 years	Attractiveness ratings and attributions for facial photographs of children w/bilateral and unilateral cleft lip vs noncleft children	Children w/clefts were viewed significantly more negatively than noncleft children in several domains (eg, children w/clefts were rated as boring, ugly, dirty, mean, weak, bad, frightened, stupid, careless). Children w/bilateral clefts were rated more negatively than those w/unilateral clefts in several domains
Tobiason (1987)	317 child raters ages 8–16 years	Attractiveness ratings and attributions for photographs of children w/ unilateral or bilateral CLP vs photographs of the same children in which the facial deformity was disguised	In the uncorrected photographs, children w/clefts were rated as less friendly, less popular, less intelligent, and less attractive than in corrected photographs
Krueckeberg et al. (1993)	Children w/ heterogeneous CFAs ($N = 30$; CLO $n = 7$, CLP $n = 5$, CPO $n = 8$) and healthy controls ($n = 22$)	Parent, teacher, and child self-report social skills; facial encoding/ decoding; and social knowledge interview	No differences between parent and teacher reported-social skills for children w/CFAs vs controls. Girls w/CFAs reported greater social competence than control girls and boys w/CFAs. No differences in facial encoding/decoding skills. In social knowledge interview, children w/CFAs provided fewer "friendly" responses than controls. No differences on dependent variables for children w/"visible" vs "invisible" CFAs

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Table 5. Studies of peer relations and social perception*(*Continued*)

Authors	Sample	Domains assessed	Results
Berry et al. (1997)	Children w/CLP ($n = 14$), CPO ($n = 6$), and matched controls ($n = 16$)	Adult ratings of personality attributes based on speech samples	No differences between personality ratings given to children with clefts vs controls
Slifer et al. (2004)	Children w/CLP ($n = 23$), CPO ($n = 5$), CLO ($n = 6$), and healthy controls ($n = 34$)	Parent-rated social competence; child self-report of social acceptance and physical attractiveness; child and parent ratings of dissatisfaction w/facial appearance; and direct behavioral observation of social interaction w/an unfamiliar peer	Parents rated children w/clefts as less socially competent than controls. No differences in child self-reports of social acceptance and physical appearance. Children w/clefts and their parents reported greater dissatisfaction w/child's facial appearance. In observed interactions, children w/clefts were less likely to make an activity choice (eg, selects an activity when presented w/a choice by their peer) and were more likely to fail to respond to a peer's direct question

*Abbreviations are explained in the footnote to Table 4.

THE CHILD-INFANT DEVELOPMENT PROJECT

Description of study and aims

In an effort to extend the findings of previous studies and address their methodological shortcomings, our research group initiated a prospective longitudinal study of infants with CLP, CPO, and sagittal craniosynostosis (ie, premature fusion of skull sutures). There was no attempt in this research to manipulate medical or psychologic treatment variables, but rather to chart the developmental course and outcome of these children in the context of typically available treatments from a regional cleft-craniofacial team. Children and their families were followed from age 3 months to 7 years. We initially recruited 76 infants with one of these diagnoses and a demographically matched group of 76 "comparison" infants without known medical disorder (for a total of 152 subjects). All children were scheduled to be seen in our laboratory at 6 time points: immediately

following recruitment (about 3 months of age) and at ages 1, 2, 5, 6 and 7 years. We were able to retain roughly 75% of our original sample through age 7, for a final sample of 113 children. In the craniofacial sample, there were no differences in attrition by diagnosis. However, attrition was related to SES, particularly in the comparison group, with better retention shown by families with higher SES.

The overall goal of the child-infant development project (CIDP) was to develop a predictive model of psychosocial functioning in children with orofacial clefts, guided by developmental theories regarding the effects of the early parent-infant relationship on the subsequent functioning of the child (Speltz, Greenberg, Endriga, & Galbreath, 1994). Specific environmental and biological variables were expected to contribute to the development of the child in a multifactorial, interactive, and reciprocal fashion. Our measures were designed to assess designated *predictor* variables (eg, cleft diagnosis and severity, parental response to the cleft condition), *outcome* variables (eg, infant neurodevelopmental status, child self-perception, behavioral adjustment, school achievement), as well as hypothesized *mediating processes* (quality of child's attachment to the parent). Major variables included maternal sensitivity, infant responsiveness to the mother, and the resulting quality of the infant's attachment to the mother, all assessed through direct observation in the clinic and detailed coding of videotapes. We also assessed the neurocognitive and psychomotor status of children throughout the length of the study, as well as many of the important medical correlates and health-related processes associated with cleft diagnoses (eg, facial appearance, speech and hearing, timing of surgeries, and other treatments).

The specific aims of our research focused on the comparison of diagnostic groups on major predictor and outcome variables (eg, CLP vs controls; CLP vs CPO), change in child status and family functioning over time, and the extent to which hypothesized predictor variables were actually related to outcomes.

Summary of findings

Cognitive and motor development

The early cognitive and motor development of infants/toddlers with clefts lagged significantly behind the development of noncleft peers (Speltz et al., 2000). However, by age 5 all groups were functioning at very similar levels and they remained so at age 7 (Collett, Speltz, & Leroux, 2006a). Specifically, when these children were preschoolers and at roughly the age of school entry, we found few statistically significant differences between cleft groups, or between cleft and control groups on measures of neurocognitive functioning and academic achievement. Among children with clefts, neurocognitive functioning and academic achievement at ages 5 and 7 were predicted by early mental and motor development (eg, BSID scores on these indices) and psychosocial variables (eg, maternal sensitivity during parent-child interactions).

Attachment and quality of parent-child relationship

Quality of mother-infant attachment was *not* affected by the presence or absence of cleft lip in children with cleft palate (Speltz, Endriga, Fisher, & Mason, 1997). However, the groups showed different *patterns of change* in attachment status over time. While attachment among controls was fairly stable between ages 12 months and 5 years, the CLP group initially showed high and then decreasing levels of effective (secure) attachment, and those with CPO showed nearly the opposite trend (ie, increased rates of secure attachment over time; Maris, Endriga, Speltz, Jones, & DeKlyen, 2000). Within the cleft group, insecure attachment was predicted by a combination of 4 maternal variables: younger age, primiparity, higher levels of self-reported depression, and lower levels of observed infant feeding skill.

Feeding and physical growth

Consistent with an earlier study (Speltz, Goodell, Endriga, & Clarren, 1994), infants

with CLP scored lower than control infants on the Clarity of Cues scale from the Nursing Child Assessment of Feeding Scale (NCAFS; Sumner & Spietz, 1994) (Endriga, Speltz, Marris, & Jones, 1998). Over time, feeding cues became increasingly clear for all groups. Although no significant group differences emerged in maternal scores on the NCAFS Sensitivity to Cues scale, this variable proved to be an important predictor of other outcomes, such as security of child attachment (Endriga et al., 1998) and cognitive-developmental outcomes (Collett et al., 2006a; Speltz et al., 2000). Using weight-for-height measures at 3 and 12 months, physical growth for children with CLP and CPO was compared with growth rates established in the commonly used pediatric growth charts from the Centers for Disease Control and Prevention (Coy, Speltz, Jones, Hill, & Omnell, 2000). Although not statistically significant, children with CPO showed a slightly accelerated rate of growth relative to population norms, while those with CLP did not differ from the population average. Importantly, psychosocial variables (eg, SES, infant temperament, social support, and mother and infant NCAFS scores) contributed to the prediction of early physical growth, even after controlling for medical variables (eg, birth weight, cleft diagnosis, early health status). This did not hold true at 12 months, when psychosocial variables provided no predictive value beyond medical variables and infants' growth at age 3 months.

Effects of facial appearance

The facial appearance of infants with clefts was much less of a risk factor than we had hypothesized, at least in the first 2 years of life. Using a Q-sort procedure, a group of adults rated the facial attractiveness of infants in all groups (Coy, Speltz, & Jones, 2002; Speltz et al., 1997). As anticipated, infants with CLP were rated as least attractive, followed by infants with CPO, and control children. Contrary to our expectation, facial attractiveness ratings were *not* predictive of child attachment security at age 12 months (Speltz

et al., 1997), a finding replicated with an independent group of attractiveness raters (Coy et al., 2002). In fact, securely attached infants were rated as less attractive than insecurely attached children. This finding has prompted us to hypothesize that caregivers may perceive infants with clefts as more vulnerable, leading to heightened activation of the "attachment system" and caregiving behaviors that foster early secure attachment.

Behavioral and social adjustment

Behavioral adjustment was assessed at ages 5, 6, and 7 by parent and teacher ratings of behavior problems (Child Behavior Checklist and Teacher Report Form) and teacher ratings of social competence in the classroom (Health Resources Inventory; Gesten, 1976). The Health Resources Inventory contains scales measuring frustration tolerance, peer socialization skills, and classroom behaviors that promote learning. Preliminary analyses of these parent and teacher measures (Collett, Leroux, & Speltz, 2006b) have indicated that children with and without clefts showed nearly equivalent levels of adjustment, as indicated by group means as well as percentages of children in each group exceeding conventional clinical thresholds. One exception was the finding that teacher ratings of internalizing and externalizing behavior problems at age 6 were significantly higher for children with clefts than those without. However, by age 7 such differences were not apparent and we tentatively conclude that there is little difference in behavioral adjustment between our cases and controls during this age period.

Emotion regulation

At age 5, children were observed during a "disappointment task" to assess emotion regulation, as described by Saarni (1984) and Cole (1986). We found that children with CLP and CPO showed less expressed disappointment (through verbalizations, facial expression, mild tantrum behavior) than control peers (Endriga, Jordan, & Speltz, 2003). It was hypothesized that this may reflect

resilience in these groups, developed after early exposure to stressful life events that require regulation of negative affect (eg, anxiety regarding surgical procedures). Alternatively, this finding might reflect an overly controlled or inhibited response pattern, comparable to that described by previous authors (eg, Richman, 1978). Longitudinal analyses suggested that expressed disappointment was predicted by parenting stress at age 2 years. Expressed disappointment was also predictive of later behavioral adjustment, as measured with the CBCL.

Summary and critique of the CIDP

Overall, the CIDP study revealed few group differences between infants and young children with orofacial clefts and demographically matched controls. Differences were observed at specific points in time (eg, cognitive and motor development in infancy), in relation to specific developmental trends (eg, diverging trends in attachment security for CLP vs CPO children, differing rates of physical growth), and in a few specific domains of function (eg, emotion regulation). Notably, and in striking contrast to our expectations, we found no evidence that children with clefts were any more likely than comparison children without clefts to show problems in behavioral adjustment or academic achievement during the late preschool and early elementary school years. It is possible, of course, that most children with clefts who eventually experience problems in behavioral adjustment or academic achievement do not show them until much later in the elementary school period, when academic and socialization demands increase rather dramatically. Most previous studies finding evidence for problems involved older children (eg, Richman & Millard, 1997; Schneiderman & Auer, 1984), although nearly all of these studies lacked control of participants' demographic characteristics.

It is also important to note that although our cases and controls did not differ on IQ and academic measures, both groups of children showed relatively high levels of potential vulnerability. For example, at age 5, just over a third of our cleft group had full-scale IQ scores at or below 90 (vs 23% of the control group, a statistically nonsignificant difference; Collett et al., 2006a). At age 7, about a quarter of the cleft group had standard scores at or below 90 on measures of arithmetic and reading comprehension, with comparison group children again showing roughly equivalent levels of performance. These findings (which are notable given the relatively high SES status of both groups; see below) suggest that despite the lack of mean group differences, there is a significant minority of cleft children who may need—or would clearly benefit from—assistance in their early academic years. Perhaps the most important findings from the CIDP, therefore, are those related to cross-time, within-cleft group analyses. In several domains, our data revealed risk factors that could be used to screen infants and young children for closer monitoring and/or preventive interventions; these could be easily incorporated into their ongoing care through cleft-craniofacial teams. For example, data coded from maternal-child feeding interactions proved to be a powerful predictor of several outcomes within this group, ranging from early physical growth to attachment security. Similarly, infancy Bayley examination scores predicted later IQ at school entry. One can easily imagine interventions that would target infants and mothers at the greatest risk, providing additional support and/or specific interventions that would foster positive early relationships and optimize early learning environments.

With respect to methodological considerations, the CIDP is one of the few prospective longitudinal studies of social-emotional development in children with orofacial clefts and, to our knowledge, one of only 2 investigations to use a matched control group of noncleft peers and multiple assessment methodologies (eg, direct behavioral observations, caregiver rating scales, clinician-administered assessments; the only other study to do so was completed by Krueckeberg, Kapp-Simon, and colleagues). However, we encountered several problems that limit the interpretation of our findings and warrant further comment.

The ascertainment and retention of participants proved to be particularly difficult. As in most studies, we recruited consecutive referrals from our hospital-based program. We have since come to appreciate the variability in cleft populations seen at different hospital-based programs around the country, because of factors that are difficult/impossible to estimate (eg, regional practice parameters; over-targeting or undertargeting of rural families vs urban families, or lower SES families vs upper SES families). A much stronger approach would be a multisite study, with regionally diverse programs chosen to maximize the generalizability of findings. Despite our efforts to retain families (eg, project newsletters, birthday/special event cards, etc), we had 22% attrition over 7 years of the study, with greater attrition among low SES families. Although SES was not related to diagnosis, it is impossible to rule out attrition bias in other important respects without achieving a higher retention rate (eg, >90%). Furthermore, attrition affects the external validity of our findings, such that age 5 and 7 findings from the CIDP can only safely be generalized to higher SES families. Attrition at these later time points also limited our statistical power to detect differences, perhaps contributing to null findings. Finally, for this project we sought to include a relatively broad and comprehensive assessment battery to evaluate a variety of social-emotional and other developmental domains. In some cases (eg, cognitive developmental outcomes), this meant that the depth of our assessments in specific domains was limited and our assessments may not have been sensitive enough to detect subtle group differences.

CLINICAL IMPLICATIONS

The research reviewed in this article, including the CIDP, has several important implications for psychosocial assessments and interventions for young children with clefts and their families. The birth of a child with an orofacial cleft and subsequent diagnosis is understandably difficult for many parents. Clinically, parents report vivid recollections and strong opinions regarding the diagnostic process, often months or years after the diagnostic event. This has now been borne out in at least a few research studies attempting to better understand parents' response to the diagnostic process (eg, Byrnes et al., 2003; Strauss et al., 1995; Young et al., 2001). Inquiring about parents' recollections of the diagnostic process, their child's treatment, and their experiences in social interactions involving their child can help parents develop a narrative of their experiences, which may reduce anxiety and improve the provider-parent relationship. Parents' narratives may also help them better understand their experiences with previous providers and their perceptions and attributions of their child's behavior across multiple social situations (eg, in play groups, in the classroom setting, etc).

In observational studies, research has suggested that the caregivers of infants with clefts may be less active and engaged than the parents of control infants. When these children reach toddlerhood, there is some evidence that parents are actually more actively engaged as "teachers" during play interactions and perhaps more sensitive. Researchers have suggested that this may reflect an effort by caregivers to stimulate their child and compensate for perceived delays (eg, Wasserman et al., 1986). Some parents can be effective in this regard, helping structure their child's play to facilitate learning and development. In other cases, these interactions may become intrusive, reflecting the parents' own anxiety rather their appraisal of what the child needs to become more comfortable or more competent in a particular situation. In our work, we have found maternal sensitivity during feeding and teaching tasks to be predictive of later child outcomes. Again, this highlights the need for the inclusion of direct behavioral observation in clinical assessment. Reviewing a videotape of these interactions with the parent is often very useful clinically, as it can provide a tool for understanding parents' subjective experience of the interaction and their perceptions of their child. With these subjective experiences in mind, parents can

be coached to use facilitating strategies during interaction with their child.

The social-emotional and self-concept outcomes for children with clefts are diverse, likely reflecting myriad factors. Overall, it appears that these children are prone to more social inhibition than others and may show a passive and self-conscious pattern of interaction. Such a pattern can be understood as a self-protective effort to avoid attention and possible teasing. As these behaviors are unlikely to draw attention in the same way that externalizing behaviors do, such youngsters may avoid detection and identification of needed assistance. In a therapeutic context, coaching and rehearsal of self-assertion and responding to peer teasing may improve the psychologic outcomes of children with clefts. Such skills could easily be incorporated into a social skills intervention for those identified as having difficulty, or as a preventive intervention targeting children with clefts prior to school entry or in the early elementary school years.

Despite the many stressors associated with having a stigmatizing and chronic medical condition, the majority of young children with nonsyndromic orofacial clefts appear to show normal social-emotional development, at least in early childhood. When these children are referred for psychosocial intervention, it is often tempting to attribute all problems to the cleft condition. Such attributions may be also apparent in the reports provided by parents and teachers regarding the child. While this view might be accurate in some cases, presenting concerns are often better explained by the same contingencies and factors that lead to referral for noncleft children, with their medical condition and treatment needs merely exacerbating symptoms or heightening levels of caregiver concern.

DIRECTIONS FOR FUTURE RESEARCH

There is still need for well-controlled, prospective studies to evaluate the social-emotional functioning of children with orofacial clefts over the course of their development. Studies that facilitate the development and refinement of developmental models, helping understand the factors that contribute to resilience and vulnerability, are more likely to advance this area than cross-sectional studies comparing cleft and control groups at one point in time. Knowing whether children with clefts on average have higher or lower self-concept ratings or behavior problems than typical peers is probably of less importance than knowing what infancy or preschool factors *within* the cleft population are most predictive of later functioning. Regardless of how they compare to "normal," school-aged children with clefts will show a range of outcomes in social-emotional development and school adjustment that is potentially predictable on the basis of their functioning during the infancy and preschool years. How do we best discriminate the majority who are likely to do well from the significant minority who are likely to struggle? During their early years of life, children with clefts constituting a "captive" clinical population, as a result of their need for surgeries and other medical treatments. From a practical standpoint, therefore, there is ample opportunity for early identification and prevention of subsequent social-emotional difficulties in this high-risk population. However, there are very few research findings upon which to develop an effective early assessment battery. Findings from the CIDP tentatively suggest promising areas of assessment for predictive purposes (eg, mother-infant feeding interactions and maternal depression, particularly among primiparous mothers), but replication and extension of these findings in larger cleft samples is necessary. Given the numbers of subjects typically required to address questions of correlation and prediction in statistical models, it will likely be necessary to conduct multisite investigations. Multisite studies would also help minimize the ascertainment bias that has likely characterized many previous studies in this area.

It is also time to begin well-controlled prevention or early intervention studies in this

population. Although researchers have been hypothesizing and writing about the social-emotional vulnerabilities of the cleft population for decades, there are few intervention studies for these children or their parents. Ideally, studies would target clinically relevant risk factors and begin testing the effects of interventions to address those needs. For example, the parent coaching and review of videotapes procedure described in the foregoing "clinical implications" section could be developed into an intervention with testable effects on mother and child outcomes. In our work, we have frequently heard parents tell us that they are uncertain of what to do when they or their child is questioned about the child's appearance in public (eg, someone comments about the child's appearance in the grocery store line). Parent coaching and role-playing procedures based on evidence-based social skills and parent behavior management interventions (eg, Eyberg, 1988; Webster Stratton, 1998) could be developed to meet this need.

Another area of potentially fruitful research is the temperamental trait "inhibition to the unfamiliar," as it has been studied in the general population by Jerome Kagan and colleagues (eg, Kagan, 1997; Kagan, Snidman, & Arcus, 1992; Mullen, Snidman, & Kagan, 1993). Although it has been hypothesized that children with orofacial clefts may show an overly inhibited response style, this is based largely on research using teacher and/or caregiver-report rating scales. The observational techniques utilized by Kagan et al. may offer a more sensitive assessment of inhibition, and would capitalize on the wealth of research in this area related to the genetic and physiological correlates of inhibition. Interestingly, there is at least one study (Arcus & Kagan, 1995) suggesting that certain craniofacial characteristics are related to inhibited temperament. We are unaware of studies using this paradigm with children with clefts; however, further research with this population exploring mechanisms that provide a conceptual "link" between inhibition and facial development has the potential to significantly advance the field.

The continued use of observational measures and other techniques to assess the social status of children with clefts (eg, sociometric techniques) may help bridge the gap between the social psychologic literature on facial attractiveness and bias and the peer networks of children with clefts. As noted above, the self-reports of children with clefts may be subject to response bias and social desirability, and parents and teachers may be less aware of the nuances of the child's peer group. Using the sociometric strategies described by Dodge et al. (Asher & Dodge, 1986; Coie, Dodge, & Coppotelli, 1982; Harrist, Zaia, Bates, Dodge, & Pettit, 1997), it would be possible to determine rates of rejection versus acceptance as reported by children's peer group. Furthermore, in combination with observational techniques and rating scale measures, it would be possible to determine what variables predict acceptance versus rejection for children with clefts and to tailor interventions accordingly (Pope & Ward, 1997).

Finally, parental response to the initial diagnosis of orofacial cleft have been studied through retrospective questionnaires or interviews, often given or conducted months or years after the event. Most of these data have centered on the content of parents' verbal responses to interview questions (eg, information preferences vs recall of information provided). However, other methodologies exist. For example, the Reaction to Diagnosis Interview (Pianta & Marvin, 1992) is a very brief (<20 minute), semistructured interview used to assess parents' response to their child's diagnosis. Parents' responses and nonverbal behaviors during the interview are videotaped and later coded to assess "resolution" (ie, acceptance of their child's diagnosis and ability to move forward). Resolved status has been found to correlate with clinically meaningful child and parent outcomes, such as parent-child attachment, parenting stress, and marital satisfaction (Marvin & Pianta, 1996; Pianta, Marvin, Britner, & Borowitz, 1996; Sheeran, Marvin, & Pianta, 1997; Welch, Pianta,

Marvin, & Saft, 2000). Much of the work with the Reaction to Diagnosis Interview has focused on parents' reactions to their child's diagnosis with cerebral palsy and other neurological conditions, although there are also preliminary studies with children with CFAs (Barnett et al., 1999). Given the existing data, it may be that parents' reactions to their child's diagnosis, as assessed with this type of instrument, would provide another target for intervention. For example, interventions might be used to train clinicians to tailor their feedback to the needs and desires of families who are likely to struggle with the acceptance of their child's diagnosis.

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