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A developmental approach to mental health for children and adolescents with orofacial clefts

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Structured Abstract

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The mental health of children, adolescents, and adults with orofacial clefts has been studied extensively. Outcomes of interest have included parental adaptation, parent–child attachment, child development, intellectual and academic outcomes, behavioral adaptation, and quality of life. The literature sheds light on mental health needs and opportunities in each of these domains at various stages of development. However, this research has been limited in several respects and methodologically rigorous studies are needed to clarify the role of mental health in craniofacial team care. In particular, randomized controlled trials investigating the efficacy of psychosocial interventions tailored for this population are long overdue. Such studies have the potential to advance routine mental health care for individuals with orofacial clefts to the level of 'evidence-based care.'

Key words: cleft lip/palate; development; mental health; orofacial cleft

Introduction

The psychosocial well-being of children with orofacial clefts and their families has been a focus of research for over 50 years. Early research sought to dispel commonly held beliefs regarding an association between clefting and intellectual impairment or psychopathology. More recent studies have investigated issues such as quality of life, child self concept, the neurobiological association between development of the face and brain, and parents' response to the diagnostic process and the discovery that their child has a craniofacial anomaly. This body of research has been used to support the inclusion of mental health providers (e.g. psychologists, social workers) as part of the 'craniofacial team', a practice which is now generally accepted in major craniofacial centers and advocated by professional organizations (e.g. American Cleft Palate Craniofacial Association). In the review that follows, we will discuss the mental health issues faced by children with non-syndromic orofacial clefts and their

families at key points in development: infancy and early childhood, preschool and primary school, and adolescence and early adulthood. We will highlight principal findings from a longitudinal study of children with orofacial clefts conducted by our own group, as well as the findings of other research groups. Finally, we close with a critique of this area of research, and discussion of steps needed to advance the 'state of the art' in mental health care for children with clefts of the lip and/or palate.

The Child Infant Development Project

The Child Infant Development Project (CIDP) was initiated primarily to study early development and parent–child attachment in infants with cleft lip and palate (CLP) and cleft palate only (CPO) relative to unaffected controls. Also of interest were neuropsychological outcomes, such as children's cognitive and early academic development. Cases with CLP or CPO (n = 28, 26 respectively) and demographically matched control children without any known medical conditions (n = 69) were recruited at age 3 months. We had the opportunity to follow this cohort of children to age 7 years, with a relatively low rate of attrition (i.e. 22% over the course of the 7-year study). As one of very few prospective case–control studies, this project offers unique insight into differences and similarities between children with clefts and non-cleft peers, as well as clinically informative data regarding factors that might increase or attenuate risk for this population.

Mental health in infancy and early childhood

Mental health issues during infancy and early childhood are broadly defined, encompassing variables such as parents' adaptation to the birth of their child, the quality of parent–child relationships, and infants' attainment of early milestones. Researchers' interest in these outcomes in young children with orofacial clefts has been driven by hypotheses regarding the potential adverse effects of a visible and stigmatizing birth defect on parents' adaptation and ability to bond with their child.

There is a growing body of research regarding parents' emotional responses to having a child with an orofacial cleft. Quantitative studies using standardized measures of parent distress have produced mixed results. It does appear that a subset of parents of children with clefts experience significant symptoms of stress, depressive symptoms, and lower self-evaluations of competence [e.g. Speltz et al. (1)]. However, differences are not consistently observed, and the parents of children with clefts generally do not experience the same level of distress as the parents of children with other medical conditions [e.g. Downs Syndrome; Pelchat et al. (2)]. Descriptive and qualitative studies have explored the unique concerns of parents of children with orofacial clefts. In particular, a few researchers have studied parents' response to the 'diagnostic event' [e.g. Young et al. (3)]. These studies are clinically informative and suggest that parents have reasonable expectations that are unfortunately often not met. That is, they want providers to: 1) use technically accurate and sensitive descriptions of their child's condition (e.g. 'cleft lip and palate' instead of 'birth defect' or 'harelip'), 2) address the possibility of associated conditions (e.g. learning disabilities or mental retardation), 3) provide practical information regarding feeding, and 4) offer feedback regarding *both* normal and abnormal aspects of their child's physical exam, rather than focusing exclusively on the cleft diagnosis (3). Not surprisingly, parents' concerns and need for information evolve over time. Initial concerns focus on the pragmatics of feeding, their child's upcoming lip repair surgery, and possible embarrassment upon introducing their child to others. Later concerns include learning more about the cause(s) of their child's cleft, ongoing craniofacial care, and the risk for recurrence in future pregnancies.

Another focus of existing studies, including the CIDP, has been the quality of parent–child relationships. Again, the hypothesis for much of this work has been that these highly visible physical conditions might affect parents' response to their baby and potentially impede parent–child bonding. Overall, it *does not* appear that rates of attachment security, as measured with standardized procedures such as the Ainsworth 'Strange Situation' protocol (4), differ among mothers and children with clefts vs. unaffected controls. In fact, we found that CLP had a somewhat *protective* effect and fostered increased rates of secure attachment at age 12 months (5). Those children with the most significant impairment in facial attractiveness (as measured via objective raters) were found to have the highest rate of secure attachment. It was hypothesized

that visible clefts might contribute to maternal perceptions of vulnerability, resulting in heightened maternal responsiveness (5). Another possibility is that the regular contact that these mothers had with their child's medical providers might have led to increased perceptions of social support.

Several studies have used other coding systems to describe parent–infant interactions. This research has provided some evidence suggesting that infants with clefts and their mothers are less playful, active, and engaged than control dyads [e.g. Wasserman et al. (6)]. In the CIDP, we were particularly interested in feeding interactions, given that this is a stressful process for many mothers of children with clefts. Using the Nursing Child Assessment Satellite Training (NCAST) Feeding scale, we assessed mother–child interactions during feeding at ages 3 and 12 months. The NCAST is used to measure maternal sensitivity, child's clarity of cues, and the overall quality of feeding interactions. In contrast to our expectations, there were few group differences on this scale (7). However, we have found that scores on the NCAST are predictive of later outcomes for children with clefts. It bears mentioning that virtually all of the work on parents' response has been with mothers, and we know little about how these processes might be similar or different for fathers.

Research on infant development suggests that children with orofacial clefts lag slightly behind their peers on clinician-administered and parent-report measures of motor and mental development (8, 9). For example, children in both cleft groups of the CIDP received lower mental development scores than controls at 3, 12, and 24 months. In motor development, those with CPO scored significantly lower than controls at all three assessment points. The motor development of children with CLP was equivalent to controls at 3 months, although it was subsequently found to be significantly lower. Comparisons among cleft groups suggest that those with CPO receive the lowest scores, followed by children with CLP and CLO, who tend to score roughly within the average range. Children with clefts as well as other associated malformations appear to be at particular risk (10).

Clinical implications and recommendations

These findings suggest several clinical implications. Having a child with an orofacial cleft is of course a stressful event, likely to violate parents' expectations about having a newborn. Perhaps because of the limited sensitivity of current measures to the particular issues these families face, this parental stress may not be reflected through standardized questionnaires. Further, this stress may not rise to the level of 'clinical significance.' Nonetheless, this population does have unique needs and concerns to address. In clinical work, we have been impressed with parents' emotionally charged recollections of their early interactions with medical personnel and either positive or derogatory comments made by nurses and doctors. Recent qualitative studies suggest that this is a common observation. Such findings are a reminder both of the vulnerability of new parents and of the importance that parents assign to their child's medical providers. On a practical level, data from studies of parents' expressed desires upon diagnosis can help providers to anticipate some of the questions that parents of children with orofacial clefts might have, as well as the ways in which these questions may develop over time. These data also suggest that it may be helpful to follow the lead of parents, soliciting their initial questions and letting them know that these worries may change over time and that support will continue to be available as needed.

It is reassuring to note that, despite subtle differences in the quality of parent–infant interactions in cleft populations, the majority of these parent–child dyads establish a secure attachment. The findings of the CIDP and other research groups suggest that observations of parent–child interaction may be informative. Given that feeding is an anxiety-provoking issue for these families, it seems reasonable to incorporate a feeding observation into clinical care. In addition to providing practical information regarding feeding technique, coding systems such as the NCAST could easily be used by nursing staff to assess more subtle aspects of the interaction. These data may then be used to guide clinicians in identifying parents who need additional coaching to facilitate feeding and to promote general sensitivity to their child's cues.

Routine developmental screening for children with clefts appears warranted, particularly for those with other risk factors (e.g. associated malformations, sociodemographic risks). Such screening runs the risk of 'false positives' (i.e. incorrectly identifying children as being at risk), potentially contributing to parents' perception of child vulnerability [i.e. 'vulnerable child

syndrome' (11)]. Although this is an important issue of which to remain mindful, the risk is outweighed by the benefits of identifying concerns early and intervening. If done well, the process of completing an early childhood evaluation can serve as an opportunity to point out a child's strengths and to pique a parent's curiosity about their child's emerging skills.

Preschool and school-age

The preschool period marks several key developmental transitions. By this age, children are able to assert their independence from caregivers and make their preferences known, resulting in frequent parent-child 'negotiations.' For many, the preschool years mark an emergence into a broader social milieu and increased contact with adults and peers outside the immediate family. This also tends to be a time when parents become acutely aware of their child's early academic prospects. These trends continue into school-age, as the peer group becomes a primary focus and parents receive evaluative feedback from teachers about how their child compares with his or her peers. Research on preschool and school-age children with orofacial clefts has focused heavily on assessments of parent–child interaction, behavioral adaptation, self-concept, and cognitive and academic functioning.

Interestingly, there is some indication from observational studies of children with clefts that parent–child interactions may differ for preschoolers vs. infants. In particular, mothers in these dyads have been found to be more actively engaged than control mothers, utilizing a high rate of teaching and behavioral commands [e.g. Allen et al. (12)]. Overall, the impression is of a mother seeking to elicit her child's best performance, sometimes in an intrusive and controlling manner. It has been speculated that this might reflect mothers' responses to their child's real or perceived developmental delays (12).

Although the construct of 'attachment' differs at this age as compared with toddlers, it continues to be informative among preschoolers. In the CIDP, there were few group differences in rates of secure attachment when children were preschool age. We did, however, find some interesting differences in the *stability* of attachment security over time. While attachment between the 12-month visit and the 5-year visit remained fairly stable among controls, rates of secure attachment among cases with CLP declined over time. Interestingly, cases with CPO showed the opposite pattern: their rates of secure attachment increased. Speltz et al. (5) speculated that facial disfigurement might elicit a protective response from mothers that facilitates attachment. It follows that this effect might decrease over time, as children's facial appearance normalizes with surgery and development.

Self-concept has been of particular interest for researchers of this age group, given the well-known social stigma associated with craniofacial anomalies. A few studies have shown that self-concept scores are lower among children with clefts relative to test norms, with particular concerns related to their physical appearance [e.g. Broder et al. (13)]. Contrary to expectations, several other studies find no such differences; in them, self-concept scores of children with clefts are as high as or even higher than those of test norms and control peers [e.g. Leonard et al. (14)].

Studies of behavioral adaptation have primarily utilized parent or teacher report measures to assess child well-being, though a few observational studies also exist. One of the more consistently reported findings, primarily from the work of Lynn Richman and colleagues [e.g. Richman & Millard (15)], is that children with orofacial clefts are rated as being more socially inhibited than their peers. It has been speculated that this may reflect a self-defense mechanism, in which children with orofacial clefts avoid scrutiny by not drawing attention to themselves. However, other studies have found few group differences between cases and controls on parent and teacher measures of behavior problems, though a sizeable minority of children with clefts may still score within the 'clinical' range [e.g. Speltz et al. (16)]. For example, in the CIDP there were few group differences between cases and controls on parent and teacher report forms of the commonly used Child Behavior Checklist (CBCL). There are a few observational studies which support the notion that of children with clefts are more inhibited than their peers. Children in the CIDP were presented with a disappointment task and their responses were coded (17). Those with CLP or CPO were less likely to verbally or nonverbally express their disappointment than control children. While this might reflect a form of resilience (i.e. effectively coping with disappointment), it may also indicate an overly

controlled or inhibited response pattern, as described by Richman et al. Other observational studies of social interaction indicate that while children with orofacial clefts have adequate conversational skills, they are less assertive than their non-cleft peers and less responsive to their peers' overtures (18).

With the entry into preschool and school settings, cognitive and early academic outcomes become key issues. There are numerous studies of cognitive development in children with clefts, primarily among those of elementary school age. Findings generally suggest that individuals with clefts score within the average range relative to test norms and comparably to their non-cleft peers on traditional measures of intellectual ability (19, 20). The CIDP and several other studies have found that verbally mediated skills (e.g. verbal reasoning, verbal learning and memory) are a relative weakness (8, 20).

Findings related to academic functioning are more striking. Richman et al. have reported notable elevations in the rate of learning disabilities among children with clefts. While the base rate of learning disabilities is estimated to be roughly 10–20% among otherwise healthy children (21), studies have reported evidence for learning disabilities in 30–40% of children with clefts, with particular vulnerability in reading (22). Further, Richman and colleagues have suggested that children with clefts who struggle with reading have a unique profile, differing from that of non-cleft children with dyslexia. Specifically, Richman & Ryan (23) have reported that children with clefts tend to have particular deficits in rapid naming, rather than the phonological deficits typically observed among non-cleft children with developmental reading disorders. As discussed below, there are some recent data from studies with adults with orofacial clefts which suggest a neurobiological foundation for such differences.

Clinical implications and recommendations

Studies of preschool and school-age children with clefts are similar to those of infants in that, though subtle differences between cleft and non-cleft dyads are observed in parent–child interactions, differences in rate of attachment security have not been observed. To some extent, parents' responses may reflect lingering anxiety regarding the possibility of developmental delays associated with their child's cleft or a need to 'compensate' for their child's condition by eliciting performance. It may also be that protective behaviors that were adaptive when their child was an infant become less so as their child begins to assert increased independence. Clinically, there may be a role for mental health providers in coaching parents in 'child-directed interaction' (CDI). Though CDI is typically used in the treatment of disruptive behavior disorders, in this context it may be used to assist parents in following their child's lead and utilizing a less directive style of interaction, thus fostering warm parent–child relationships.

There is also some indication that social skills intervention might benefit children with clefts in this age group. Target skills might include dealing with appearance-based teasing, initiating social interactions, and assertiveness. Kapp-Simon and Simon (24) have a social skills curriculum for children with craniofacial anomalies, as well as preliminary evidence for efficacy among adolescents (25). Inclusion of parents in social skills intervention may also be helpful. For example, parents might be coached in the use of adaptive responses to comments their child may receive from peers – or even adults – in social and community settings.

Early educational screening is one of the more promising areas for mental health involvement, and particularly relevant at preschool and early elementary school age. Research on reading in non-cleft children has progressed immensely in the last 10 years. There are data showing that reading disabilities can be identified based on precursors of reading (e.g. phonological awareness) as early as the preschool years (26). Longitudinal data suggest that reading disabilities that persist into the second grade of elementary school are very likely to continue in the absence of intervention [e.g. Fancis et al. (27)]. Universal screening for early reading problems may eventually become routine, much like newborn hearing screening in many states. Though additional data are needed to clarify the prevalence and phenomenology of reading problems among individuals with orofacial clefts, monitoring of early academic development and advocacy for early intervention appear warranted.

Adolescence and early adulthood

It is well known that adolescence is a particularly turbulent developmental period. Teenagers typically

struggle with puberty, identity issues, peer acceptance, conformity to peer group mores, and ultimately, emancipation from parents and family. Most clinicians believe that adolescence is particularly difficult for those with CLP, due primarily to factors and processes associated with their facial appearance. These include the integration of facial differences into an already changing and uncertain body image, desire for and development of romantic relationships despite dissatisfaction with one's own facial appearance, and coping with surgeries that may alter facial appearances, but are unlikely to eliminate facial scarring (28). Adolescence is also a time when parents and teenagers may intensely disagree about what constitutes the 'endpoint' of surgical treatment (28). Often, the adolescent insists that he or she does not want further surgery, but parents are committed to a long-term course of treatment that includes one or more revisions of facial scarring. The parents' call for more surgery may be interpreted by the sensitive teen as implicit criticism of her current appearance. To further complicate matters, much of this discussion may occur amidst the teen's struggle to relate to medical staff as a young adult rather than as a child.

In some cases, strong resistance from the 'over-protective' parent, and/or ambivalence on the part of the highly dependent adolescent, may complicate the adolescent's emancipation from the family. Conflict or delay in the emancipation process could theoretically hinder the individual's transition to independent adult life, which includes the development of romantic partnerships and the initiation of vocational objectives.

The extent to which these anticipated problem areas translate into actual conflict or dysfunction is unclear, as there have been few studies of the psychological adjustment of adolescents with CLP or CPO. It is known that, among school-age children nearing adolescence, children with clefts show higher than expected levels of internalizing behavior problems (as reported by parents and teachers) and, depending on age and gender, higher externalizing problems as well. For example, Richman & Millard (15) found that girls with CLP had normal levels of externalizing behavior problems until reaching pre-adolescence (ages 11 and 12), at which point they received externalizing behavior problems scores nearly 3 standard deviations above the mean. Increasing concern with appearance and negative self-appraisal were the suspected causes of this change in functioning. Studies of self concept in adolescents with clefts have also shown that older girls generate lower scores on a variety of measures related to popularity, unhappiness and satisfaction with facial appearance (13, 14).

Many young adolescents with facial anomalies, including CLP, appear to have specific social skill deficits. In one study, these individuals were observed to make fewer social overtures to their peers at school than typical children and, in turn, were approached less frequently by peers (29). Moreover, they were more likely than typical peers to engage in ineffective approach behaviors.

More recent studies utilizing 'quality-of-life' (QOL) interviews have compared the responses of adolescents with and without visible facial disfigurement (mostly CLP). In one such study, adolescents with facial anomalies reported significantly lower QOL than did the control group, with scores adjusted for age, gender, and depressive symptomatology (30). One exception to this overall pattern was found in the area of family relationships, in which adolescents with facial anomalies reported *higher* average QOL than did the comparison group. This finding was attributed to parental over-protectiveness and age-excessive levels of adolescent dependence on the parent, presumably resulting from many years of elevated parental care of the child.

The psychological adjustment of adults with CLP/CPO has been relatively well studied, with investigations dating back to the early 1970s. Nearly all of these studies, including several with comparison groups [e.g. Peter & Chinsky (31)], found that while individuals with CLP/CPO were functioning well from a psychiatric perspective, they were more likely than peers to encounter interpersonal difficulties. Indicators of these difficulties include lower rates of participation in social activities and community organizations, an older average age of marriage, and lower levels of marital satisfaction. As with adolescents, more recent research in this area has tended to employ QOL surveys and interviews [e.g. Sinko et al. (32)]. Most have found that adults with CLP/CPO report lower QOL and higher levels of social distress than comparison groups. Sinko et al. (32) found that most QOL scales in their study (e.g. 'social functioning') were correlated with individuals' level of satisfaction with their facial appearance (i.e. higher QOL associated with higher appearance

satisfaction and less desire for further surgical treatment).

Although intervention data are again limited, there have been a few studies with adolescents and young adults. Two areas have received particular emphasis (28): 1) parent–adolescent discussion and resolution of conflicts regarding surgical reconstruction; and 2) social competence training for the inhibited/shy adolescent. With respect to surgical reconstruction, a useful paradigm to assist families in identifying and articulating their positions is the Self-Understanding Model (24). In it, participants are asked to identify the thoughts, feelings, physiological reactions and behaviors typically associated with their interactions and decision-making processes regarding surgical issues. Parents and teenagers are then coached to interact more effectively when discussing these issues.

With respect to social competence, Kapp-Simon (28) has also detailed a program for teenagers with special needs. This program addresses five basic categories of social skill: 1) social initiation, 2) conversational skills, 3) assertion or direct communication, 4) empathy or active listening, and 5) conflict resolution and problem solving. Likewise, Nicola Rumsey and colleagues have embarked upon a program of research in which they have developed tools that measure adolescents' self-reports of appearance-related social difficulties and competencies. They have also developed 'Changing Faces,' a very promising intervention that teaches non-confrontational coping strategies to children and adolescents ages 11–19. After discovering that about 75% of 11- to 13-year olds with facial differences reported teasing or bullying about their appearance, Lovegrove & Rumsey (33) compared the Changing Faces intervention to a non-intervention control group. Six months after the intervention, participating teens reported a two-thirds reduction in bullying (in comparison with pre-intervention levels), improved global self-esteem, and greater confidence in handling teasing and bullying. The non-intervention group did not show these changes. These findings are promising, but require replication with more rigorous methodology including random assignment of participants to intervention and use of a placebo–control comparison group.

Published reports of interventions for adults with facial disfigurement, including CLP, are rare, despite the widely recognized influence of psychological factors on adults' desire to pursue corrective surgery [e.g. Sinko et al (32)]. The effectiveness of cognitive behavior therapy (CBT) to reduce social anxiety and appearance-related distress has been tentatively shown in one study of adults with severe facial disfigurement (34). Referred patients reported significant pre- to post-treatment increases in positive affect and life satisfaction and reductions in the self-perceived conspicuousness of their condition (there was no control group). The extent to which CBT might have similar effects on adults with less severe conditions such as CLP has yet to be demonstrated, but appears promising.

A final set of studies that deserve mention is the recent work by Nopolous et al. [e.g. (35, 36)] at the University of Iowa. In a sample of adult males with clefts and demographically matched non-cleft controls, Nopolous et al. have examined structural brain differences using MRI. Findings have indicated that males with clefts have cortical and other brain anomalies apparent on MRI (e.g. decreased cerebellar, temporal lobe, and posterior cerebral volumes; increased anterior cerebral volume). Further, they have correlated these anomalies with performance on neuropsychological measures, showing that structural abnormality is associated with poorer performance. Interestingly, they have also found correlations between structural brain anomalies and men's social functioning (36). Using data from a subset of men drawn from this cohort, Goldsberry et al. (37) recently found that males with CLP showed abnormal brain activation on PET scans during complex reading tasks (e.g. hypoactivity in left middle temporal gyrus, right medial superior posterior cerebellum; hyperactivity in a cortical–thalamic–cerebellar circuit). The authors posit that this finding represents neural inefficiency, with under-activation of areas thought to be important for language processing and increased (compensatory) activation of other regions. Although this work warrants replication by other research groups, it is intriguing and suggests that some of the neuropsychological and psychosocial features reported in earlier literature may have a neurobiological basis.

Clinical implications and recommendations

Although adolescence brings a number of challenges, there are also several clinical opportunities that emerge during this developmental period. For example, while

information gathering at earlier ages often relies heavily on parent report, teenagers are able to report on their own experience, including their satisfaction with treatment and their psychosocial concerns. Clinicians can support this transition by increasingly directing questions and comments to the adolescent rather than to the parent, and by responding to any concerns raised by the teen. Often, this shift requires persistence – an intrusive parent might reflexively answer questions directed toward their child or even openly discount those concerns that the adolescent is brave enough to raise. Although medical decisions will not be left entirely to a young adolescent, the patient's long-term interests (e.g. engagement in his or her own medical care) are best served by active and engaged participation.

Adolescence is a time of vulnerability, and clinicians may observe mental health concerns during this period even in a child who was previously known to be robust and resilient. Certainly, available data show that continued screening for concerns regarding self-concept, quality of life, and social adaptation is warranted. Developing a system for mental health screening for adolescents and young adults with orofacial clefts (e.g. using standardized rating scales) may be helpful in identifying concerns and may ultimately reduce the stigma associated with talking about psychosocial issues. Encouragingly, there are some preliminary data on psychosocial interventions for this age group [e.g. Kapp-Simon et al. (25), Lovegrove & Rumsey (33)].

Finally, the recent studies by Nopoulos et al. are among the more exciting advancements in the literature. At present, these are more useful in understanding the etiology of some of the psychosocial and neurocognitive findings than they are in directing clinical care. Ultimately, however, insight into the neurobiological similarities and differences between the cleft population and others (e.g. those with developmental dyslexia) could help to guide intervention efforts.

Research critique and future directions

In many respects, research in the area of mental health among children and adolescents with orofacial clefts can be considered a success story. Clinicians and researchers identified the need for research in this area relatively early (before the importance of psychosocial factors was recognized for many other pediatric conditions), and the medical community seems to acknowledge that psychosocial issues are important and that mental health has a role in comprehensive care. However, progress in this area of research has, over the past decade, been disappointingly limited. A noteworthy example is that published research continues to be primarily descriptive, most often comparing the scores of a small sample of individuals with clefts with test norms on a given measure. Studies including a control sample and studies utilizing a longitudinal design are very limited. Further, the methodology and methodological quality of studies are so varied that it is difficult to compare findings across studies. In their recent review of this literature, Hunt et al. (38) identified 652 abstracts related to psychosocial outcome for individuals with non-syndromic CLP. Of these, 64 (i.e. <10%) were determined to be suitable for review based on basic quality indicators, only 30 included a control sample, and the vast majority were cross-sectional (vs. longitudinal). *Most notably, despite all of the research on the mental health needs of children with orofacial clefts, we are not aware of a single randomized control trial of a psychosocial intervention*. Why is this the case, and what are the implications for clinical care and the future of mental health care for children with clefts?

Research challenges

In our work, we have encountered several challenges that likely pertain to other research groups as well. The first has to do with the recruitment of cases and healthy control samples. The decision to participate in psychosocial research is an interesting one, and by virtue of their willingness and ability to take part there may be important differences between participants and non-participants. Motivations might include a benevolent desire to contribute to research, perceived benefits of obtaining a psychosocial and/or developmental assessment for one's child, a financial incentive (if the study involves compensation), or (for cases, in particular) a desire to please one's medical providers. Any of these may be problematic, often in subtle ways that are difficult to measure. For example, families who are motivated to have their child evaluated may have concerns about his/her development and seek a research evaluation as a low-cost and

non-threatening solution. Those parents who are interested in research and who have the time and ability to participate for benevolent reasons may differ from the population in other meaningful ways (e.g. perhaps greater education, higher SES, higher level of intellectual curiosity). These issues are compounded in a longitudinal study, which involves a significant time commitment for assessment visits, often over a period of several years. Those participants who remain in the study throughout its duration may differ from others in obvious ways (e.g. more stable geographic location), as well less obvious ways (e.g. ongoing concern about their child's development). Those participants who we would most want to retain and learn more about are often the most elusive. For example, although attrition was relatively low overall in the CIDP, we had steadily declining numbers of lower income families in both case and control groups. Thus, by age 7, the findings are most applicable to middle and upper income families.

Given the difficulties of recruitment and the somewhat low base rate of orofacial clefting in the population much of this research has involved heterogeneous groups of children that include participants with other craniofacial anomalies (e.g. craniosynostosis, hemifacial microsomia). Because of limited sample sizes, researchers often collapse across cleft groups (i.e. including both CPO and CLP). Similarly, other relevant sub-group analyses (e.g. by sex, SES) are often precluded due to small sample sizes. Such limitations affected our own work with the CIDP, particularly with attrition by age 7. These issues may, in part, help to account for the diverse findings in the field. Specifically, it is difficult to recruit a sample large enough to achieve statistical power to detect differences, particularly when those differences are likely to be subtle (e.g. a 10-point difference in verbal IQ). Sampling differences from one study to the next may 'wash out' any salient effects. Large multi-site studies are an obvious solution to many of these challenges, allowing for the recruitment of larger and more diverse samples. In addition to allowing for the investigation of subtle effects, or subgroup differences, such studies are more likely to produce meaningful negative findings. That is, a failure to find a difference is more likely to be considered meaningful if it cannot be attributed to a lack of statistical power or characteristics unique to the patient population at a given site.

Psychosocial intervention and prevention studies bear their own unique problems. As most craniofacial centers are housed within tertiary care centers, they often have a broad catchment area. As challenging as it is to recruit participants for descriptive studies, it is even more difficult to bring participants in for multiple intervention and assessment visits. Relative to other pediatric populations (e.g. pediatric oncology patients), those with clefts have less frequent hospital visits, making access even more difficult. Taking the intervention into the community in the form of home visits or school-based intervention is another option, though this also presents feasibility issues (e.g. staff travel time).

Implications

Mental health care providers for children with orofacial clefts find themselves in a precarious position. Although several potential mental health needs have been identified in this group, data are mixed and one might reasonably question the evidence in support of mental health involvement. Is involvement needed because the parents of children with clefts have considerable difficulty coping with their child's condition? A percentage certainly do, but this has neither been observed consistently nor found to be a substantial detriment to either child or parent. Is it because a cleft is socially stigmatizing and has an adverse effect on self-esteem and social functioning? There are some data to suggest that this is the case, though again findings are very mixed with some studies suggesting that individuals with clefts have even higher self-esteem than non-cleft peers. Is mental health involvement needed because youth with clefts are likely to be socially inhibited and have an overly controlled interpersonal style? Although this is one of the more consistently reported findings, data are mixed and the degree of functional impairment associated with these interpersonal features is unclear. Perhaps more to the point, *in the absence of well-designed psychosocial intervention studies, what is the evidence that we are helping with any of these issues?*

If these questions have not already been asked by craniofacial providers, they probably should be. To be clear, we are not arguing against psychosocial involvement in craniofacial care and, as psychologists who work closely with a craniofacial team, we do believe that there are many valid reasons to continue to

include psychologists and other mental health providers as part craniofacial team care. However, additional work can and should be carried out to support this model, in order that we may ensure that we are providing care that is both necessary and effective. There are several areas where this may be viable – reading is a prime example.

There are now several studies suggesting that children with clefts are more likely than their peers to be referred for special education and to struggle with reading in particular. Estimates suggest that just over one third of children with clefts exhibit a reading disability, which would make them two to three times more likely to have a reading disorder than their non-cleft peers. The literature on reading disabilities in otherwise healthy children is striking. In the absence of intervention, continuity is quite high, with sequelae including a higher rate of behavior problems, school attrition, and poor employment outcomes (39). This makes reading among individuals with clefts a profoundly important public health issue. However, further studies are needed in order to: 1) determine the prevalence of reading problems in individuals with clefts using a rigorous epidemiological approach (e.g. multi-site studies including demographically matched controls), 2) establish the adequacy of existing systems in identifying and addressing early school problems among those with clefts, and 3) determine the efficacy of existing intervention models for the reading problems observed in this population. Assuming that the rate of reading problems remains high when evaluated using these more rigorous approaches, the modest expense of routine screening is certainly justifiable. Such screening could result in the identification of reading problems prior to school entry, allowing for intervention when it is most likely to be effective. Clinically, our impression has been that existing systems for identifying early learning problems do not fully address the needs of many children with clefts. Furthermore, the parents of children with clefts report that these programs are less than adequate. Often, even when a child has been evaluated to determine his or her eligibility for additional educational support, the findings are conveyed to parents in a manner that they find incomprehensible. Alternately, a family's visit with the craniofacial team could be an excellent venue to discuss a child's early educational development, given the ongoing relationship that exists between parents and this group of providers. Finally, we are not aware of studies investigating the efficacy of reading intervention and prevention programs for children with clefts. The possibility that the nature of reading problems differs in this population suggests that existing intervention models may not be applicable or effective in the cleft population. Again, additional data on this issue could help to justify routine educational screening by psychologists or other members of the craniofacial team who have expertise regarding the unique needs of children with clefts.

As discussed above, there are several other psychosocial outcomes for which screening and involvement may be justified with additional data, including areas such as parental coping, and child social skills and problem solving. To be most useful, studies of these and other outcomes will need to: 1) include multiple sites, both to recruit a sufficient number of participants to investigate sub-group differences and potential moderators/mediators of outcome as well as to illuminate any potential differences by region or care setting; 2) include demographically comparable control samples for comparison; 3) utilize well-validated measures that allow for comparisons with existing literature, ideally using a multi-method, multi-informant approach; 4) use longitudinal designs to evaluate developmental trends; and 5) investigate the efficacy of psychosocial interventions. Further, the goals of this work ought to be to investigate the full spectrum of functioning, including areas of strength and weakness, and to optimize functioning rather than just ameliorating deficits.

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References

1. Speltz ML, Armsden GC, Clarren SS. Effects of craniofacial birth defects on maternal functioning postinfancy. *J Ped Psych* 1990;15:177–96.
2. Pelchat D, Ricard N, Bouchard JM, Perreault M, Saucier JF, Berthiaume M et al. Adaptation of parents in relation to their 6-month old infant's type of disability. *Child: Care, Health, & Dev* 1999;25:377–97.
3. Young JL, O'Riordan M, Goldstein JA, Robin NH. What information do parents of newborns with cleft lip, palate, or both want to know? *Cleft Palate Craniofac J* 2001;38:55–58.

4. Ainsworth MDS, Blehar MC, Waters E, Wall S. *Patterns of attachment: A psychological study of the Strange Situation*. Hillsdale, NJ: Lawrence Erlbaum Associates, Inc; 1978.
5. Speltz ML, Endriga MC, Fisher PA, Mason CA. Early predictors of attachment in infants with cleft lip and/or palate. *Child Dev* 1997;68:12–25.
6. Wasserman G, Allen R, Solomon R. Limit setting in mothers of toddlers with physical anomalies. *Rehab Lit* 1986;47:290–4.
7. Endriga MC, Speltz ML, Maris CL, Karen J. Feeding and attachment in infants with and without orofacial clefts. *Infant Behav Dev* 1998;21:699–712.
8. Jocelyn LJ, Penko MA, Rode HL. Cognition, communication, and hearing in young children with cleft lip and palate and in control children: a longitudinal study. *Pediatrics* 1996;97:529–34.
9. Speltz ML, Endriga MC, Hill S, Maris CL, Jones K, Omnell ML. Cognitive and psychomotor development of infants with orofacial clefts. *J Ped Psych* 2000;25:185–90.
10. Swanenburg De Veye HFN, Beemer FA, Mellenbergh GJ, Wolters WHG, Heineman-De Boer JA. An investigation of the relationship between associated congenital malformations and the mental and psychomotor development of children with clefts. *Cleft Palate Craniofac J* 2003;40:297–303.
11. Green M, Solnit AJ. Reactions to the threatened loss of a child: A vulnerable child syndrome. Pediatric management of the dying child, part III. *Pediatrics* 1964;34:58–66.
12. Allen R, Wasserman GA, Seidman S. Children with congenital anomalies: the preschool period. *J Ped Psych* 1990;15:327–45.
13. Broder HL, Smith FB, Strauss RP. Effects of visible and invisible orofacial defects on self-perception and adjustment across developmental eras and gender. *Cleft Palate Craniofac J* 1994;31:429–36.
14. Leonard BJ, Brust JD, Abrahams G, Sielaff B. Self-concept of children and adolescents with cleft lip and/or palate. *Cleft Palate Craniofac J* 1991;28:347–53.
15. Richman LC, Millard T. Brief report: Cleft lip and palate: Longitudinal behavior and relationships of cleft conditions to behavior and achievement. *J Ped Psych* 1997;4:487–94.
16. Speltz ML, Morton K, Goodell EW, Clarren SS. Psychological functioning of children with craniofacial anomalies and their mothers: follow-up from late infancy to school entry. *Cleft Palate Craniofac J* 1993;30:482–9.
17. Endriga MC, Jordan JR, Speltz ML. Emotion self-regulation in preschool-aged children with and without orofacial clefts. *J Dev Beh Peds* 2003;24:336–44.
18. Slifer KJ, Amari A, Diver T, Hilley L, Beck M, Kane A et al. Social interaction patters of children and adolescents with and without oral clefts during a videotaped analogue encounter. *Cleft Palate Craniofac J* 2004;41:175–84.
19. Hubbard TW, Paradise JL, McWilliams BJ, Elster BA, Taylor FH. Consequences of unremitting middle-ear disease in early life: otologic, audiologic, and developmental findings in children with cleft palate. *New Engl J Med* 1985;312:1529–34.
20. Richman LC. Cognitive patterns and learning disabilities in cleft palate children with verbal deficits. *J Speech Hearing Res* 1980;23:447–56.
21. Rutter M, Caspi A, Fergusson D, Horwood LJ, Goodman R, Maughan B et al. Sex differences in developmental reading disability: new findings from 4 epidemiological studies. *JAMA* 2004;291:2007–12.
22. Broder HL, Richman LC, Matheson PB. Learning disability, school achievement, and grade retention among children with cleft: a two-center study. *Cleft Palate Craniofac J* 1998; 35:127–31.
23. Richman LC, Ryan SM. Do the reading disabilities of children with cleft fit into current models of developmental dyslexia? *Cleft Palate Craniofac J* 2003;40:154–7.
24. Kapp-Simon KA, Simon DJ. *Meeting the Challenge: A Social Skills Training Program for Adolescents with Special Needs*. Chicago, IL: University of Illinois at Chicago Press; 1991.
25. Kapp-Simon KA, McGuire DE, Long BC, Simon DJ. Addressing quality of life issues in adolescents: social skills interventions. *Cleft Palate Craniofac J* 2005;42:45–50.
26. Lonigan CJ. Development, assessment, and promotion of pre-literacy skills. *Early Educ Dev* 2006;17:91–114.
27. Francis DJ, Shaywitz SE, Stuebing KK, Shaywitz BA, Fletcher JM. Developmental lag versus deficit models of reading disability: a longitudinal individual growth curve analysis. *J Educ Psych* 1996;88:3–17.
28. Kapp-Simon K. Psychological interventions for the adolescent with cleft lip and palate. *Cleft Palate Craniofac J* 1995;32:104–8.
29. Kapp-Simon KA, McGuire DE. Observed social interaction patterns in adolescents with and without craniofacial conditions. *Cleft Palate Craniofac J* 1997;34:380–4.
30. Topolski TD, Edwards TC, Patrick DL. Quality of life: how do adolescents with facial differences compare with other adolescents? *Cleft Palate Craniofac J* 2005;42:25–32.
31. Peter JP, Chinsky RR. Sociological aspects of cleft palate Adults: I. Marriage. *Cleft Palate J* 1974;11:259–309.
32. Sinko K, Jagsch R, Prechtl V, Watzinger F, Hollmann K, Baumann A. Evaluation of esthetic, functional, and quality-of-life outcome in adult cleft lip and palate patients. *Cleft Palate Craniofac J* 2005;42:355–61.
33. Lovegrove E, Rumsey N. Ignoring it doesn't make it go away: adolescence, appearance, and bullying. *Cleft Palate Craniofac J* 2005;42:33–44.
34. Kleve L, Rumsey N, Wyn-Williams M, White P. The effectiveness of cognitive-behavioural interventions provided at Outlook: a disfigurement support unit. *J Eval Clin Pract* 2002;8:387–95.
35. Nopoulos P, Berg S, Canady J, Richman L, Van Demark D, Andreasen NC. Structural brain abnormalities in adult males with clefts of the lip and/or palate. *Genet Med* 2002;4:1–9.
36. Nopoulos P, Choe I, Berg S, Van Demark D, Canady J, Richman L. Ventral frontal cortex morphology in adult males with isolated orofacial clefts: relationship to abnormalities in social function. *Cleft Palate Craniofac J* 2005;42:138–44.
37. Goldsberry G, O'Leary D, Hichwa R, Nopoulos P. Functional abnormalities in the neural circuitry nonsyndromic clefts of the lip or palate. *Cleft Palate Craniofac J* 2006;43: 683–90.
38. Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. *Eur J Orthod* 2005;27:274–85.
39. Rutter M, Kim-Cohen J, Maughan B. Continuities and discontinuities in psychopathology between childhood and adult life. *J Child Psychol Psychiatry* 2006;47:276–95.