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Neuropsychological, Behavioral, and Academic Sequelae of Cleft: Early Developmental, School Age, and Adolescent/Young Adult Outcomes

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Abstract

This article reviews behavioral, neuropsychological, and academic outcomes of individuals with cleft across three age levels: 1) infancy/early development, 2) school age, and 3) adolescence/young adulthood. The review points out that attachment, neurocognitive functioning, academic performance/learning, and adjustment outcomes are the result of a complex interaction between biological and environmental factors and vary with developmental level, sex, and craniofacial anomaly diagnosis. The degree to which associated genetic or neurodevelopmental conditions may explain inconsistent findings is unknown and suggests the need for caution in generalizing from group data on cleft.

Keywords

cleft lip and palate; craniofacial; neuropsychological; behavior; learning; adjustment

Children with cleft lip and/or palate (CL/P) are at risk for psychological difficulties due to multiple factors associated with the condition. Speech problems and facial differences may create environmental reactions from others, which may place psychological stress on the child resulting in issues with self-esteem and emotional development (Kapp-Simon et al., 1992; Richman, 1983; Millard and Richman, 2001). Furthermore, others' responses may also result in a negative behavioral response from the affected child (Pope and Ward, 1997; Richman and Millard, 1997). For many children with cleft, delayed speech and language development has been shown to be related to subtle auditory memory deficits, expressive language difficulty, and sometimes more severe central language impairment, which can result in long-term reading deficits (Richman, 1980) and other learning disabilities (LD; Richman et al., 2005).

Recent research has shown that there are neuropsychological patterns of specific strengths and weaknesses associated with clefting and some of these have been related to direct

neuromaturation differences through brain imaging (Conrad et al., 2009; Nopoulos et al., 2007). Although the early literature tended to give the behavioral, learning, and neuropsychological characteristics mentioned above more of an environmental explanation, more recent studies have been demonstrating a greater relation to biologically-based neuronal differences for some children with cleft (Conrad et al., 2009; Nopoulos et al., 2007). This paper will review the neuropsychological, behavioral, and learning/academic aspects of research on CL/P. The review will present the findings by early development, school age maturation, and adolescent/adult groups.

Psychosocial and Behavioral Aspects of Cleft

Early Development

Parents of children with congenital conditions such as CL/P often experience unique challenges during the neonatal period for a variety of reasons that may include but are not limited to: ambiguity with regard to the diagnosis and prognosis; emotional reactions and adjustments to the child's physical differences (Brantley and Clifford, 1980; Drotar et al., 1975) and subsequent management of their children's medical care. Multidisciplinary management of CL/P, which typically begins shortly after birth, often consists initially of genetic counseling, assessment, and presurgical treatment, and is followed over the next few years by surgical repair of the cleft lip, orthodontic treatment, surgical treatment and repair of the cleft palate and other forms of assessment of speech, orthodontic, and/or skeletal issues that might warrant further intervention or surgical treatment (Bardach et al., 1992). Despite ample evidence of the unique challenges faced by individuals with CL/P and their parents, the impact of these early challenges on the development of healthy infant attachment with parents and later developmental outcomes of children with cleft is unclear (Speltz et al., 1993).

A number of studies have focused on parent-child interactions, maternal characteristics, and infant characteristics as possible origins for psychosocial and behavioral problems which emerge in later development (Lyons-Ruth et al., 1990; Speltz et al., 1997). Studies have documented higher self-report ratings by mothers of children with repaired CFA of emotional distress and marital conflict and lower self-confidence in parenting skills (Speltz et al., 1990, 1993); there are also reports of less playful and engaged parent-child interactions and more controlling parent interactions (Speltz et al., 1990) in comparison to parents of children without CFA and control dyads. In general, however, few differences in mother-child attachments and interactions have been found between CL/P and control children (Speltz et al., 1993; Maris et al., 2000). In fact, some studies have reported higher rates of secure attachment (Speltz et al., 1997) in CL/P mother-infant dyads and lower-than-normal rates of psychological problems (Eiserman, 2001), which may point to the existence of protective and/or resiliency factors (Richman, 1995).

School Age

Entrance into elementary school presents important opportunities for all children to begin to acquire the self-concept and social skills necessary for navigating relationships with authority figures (e.g., teachers) and same-aged peers. The years between school entry and puberty are also characterized by advances in executive control, or emotional and behavioral regulatory skills (i.e., self-control; inhibition; self-monitoring) important for learning and navigating peer relationships (Best and Miller, 2010). Delays or deficits in self-monitoring and regulation may be more readily recognized during elementary school than infancy/toddlerhood as a function of ongoing prefrontal cerebral maturation (which continues throughout adolescence and young adulthood into the mid- to late-twenties) and corresponding increases in the demands of the child's environment. Emotional/behavioral

regulation problems may be salient for teachers, counselors, and other adults who have observed that a child's ability to attend, comply with demands, cooperate, and inhibit or delay behavioral responses (e.g., waiting to use the restroom, play, eat, etc. until bathroom breaks/recess/lunch) may be compromised in some way. Psychosocial and behavioral concerns often arise for a child with CL/P if his or her academic performance, peer relationships, behavior, and/or mood appear distinct from his or her classmates (Kapp-Simon, 1986; Leonard et al., 1991). These difficulties interact with the child's natural temperament and have the potential to affect his or her behavior, self-concept, and socio-emotional adjustment.

Parent and teacher ratings of internalizing (e.g., anxiety, depression, somatization, and social inhibition or withdrawal) and externalizing (e.g., opposition, aggression, noncompliance, and hyperactivity/impulsivity) behavior problems are significantly higher in children with CL/P than same-age peers (Millard and Richman, 2001; Hunt et al., 2005; Hunt et al., 2007; Richman and Eliason, 2008). Overall psychosocial functioning and incidence of behavioral problems do not appear to be related to the type and/or severity of cleft (Richman, 1978; Starr, 1978, 1980; Speltz et al., 1993; Richman and Millard, 1997; Millard and Richman, 2001; Hunt et al., 2005), though some evidence has suggested variation by age, gender, and visibility of cleft type (Thomas et al., 1997; Millard and Richman, 2001). For example, in a 9-year study of 44 children with CL/P who were rated annually by parents from the age of 4 to 12, Richman and Millard (1997) found that girls showed comparative levels of internalizing problems to the contrast group at 4 to 6 years of age, and significantly increased levels of internalizing problems thereafter. In contrast, boys demonstrated a mild but constant elevation in internalizing problems across all developmental levels assessed, though the increased level of parent-reported internalizing problems did not reach a level of significant pathology (Richman and Millard, 1997).

Variable rates of externalizing behavior problems in school-age children with CL/P compared to adolescents with CL/P have been reported (Hunt et al., 2007; Kapp-Simon et al., 1992; Richman and Millard, 1997; Millard and Richman, 2001). For example, Richman and Millard (1997) found that while rates of externalizing behavior problems for females were similar to the contrast group until age 11 or 12, females with CL/P demonstrated significantly increased rates of externalizing behaviors from age 13 on. Males, in contrast, exhibited increased rates of externalizing problems at ages 6 and 7, but were similar to the contrast group at ages 11 and 12. Again, the levels of externalizing behaviors did not suggest overt disruptive behavior disorder. Some conflicting findings on rates of behavior problems in school age children exist. For example, though Snyder and Pope (2010) recently found significantly lower-than-normative rates of internalizing and externalizing problems in school-aged children with CL, increased rates of behavioral inhibition have been found in a number of observational studies (e.g., Endriga et al., 2003; Slifer et al., 2004).

Adolescence and Young Adulthood

Adolescence is characterized by physical and emotional changes as well as heightened emphasis on peer relationships, social experimentation, and sexual exploration. Self-perceptions of difference from one's peer group, particularly with regard to appearance and ability, can be a significant source of distress for adolescents and may place them at greater risk for academic, behavioral, and other adjustment problems. Though the general findings on adults with CL/P do not indicate significantly increased risk for *severe* problems in psychological well-being (Clifford et al., 1972; Richman and Harper, 1980), increased levels of internalizing and externalizing behavior problems in adolescents have been reported in comparison to adolescents without cleft (Richman and Millard, 1997). Psychosocial and behavioral outcomes in groups of adolescents with cleft have been shown to vary between males and females and by age/developmental level. For example, elevations in externalizing

problems and behavioral inhibition have been found in adolescent females with CL/P and associated with increased risk for depression (Richman and Millard, 1997).

Increased rates of internalizing and externalizing behavior problems, including anxiety and depression, have been found in adolescents and young adults with CL/P (Ramstad et al., 1995). For adult males with CL/P, problems in behavioral and psychological adjustment are, in turn, related to significantly increased risk for interpersonal difficulties (e.g., decreased marital satisfaction; more infrequent participation in social or community activities; Peter and Chinsky, 1974; Peter et al., 1975); restricted social relations (Ramstad et al., 1995; Marcusson et al., 2001); and lower quality of life, which has been found to be marginally lower in adults with repaired CL/P compared to adults without cleft (Marcusson et al., 2001).

Given the saliency of issues pertaining to self-concept in adolescence and young adulthood, this section will also briefly review the findings on variables that are associated with psychological adjustment in adolescents with cleft, such as satisfaction with appearance, perceived speech problems, and use of avoidant coping strategies (Berger and Dalton, 2011). Research has suggested a significant relationship between cleft visibility and self-concept (Leonard et al., 1991; Broder et al., 1994). Findings from a study of 102 adolescent CL/P patients (ages 13–19) and their parents receiving team-based cleft care indicated elevated concerns about speech and appearance (Strauss et al., 1988). In another study, 73% of 15- and 20-year olds with CL/P reported that their self-confidence was very much affected by their cleft (Turner et al., 1998).

A more recent study by Berger and Dalton (2011) identified social experiences (i.e., frequency of social events/interactions; availability and helpfulness of parental and family social support; level of life stress present outside the parent-child relationship) as the best predictor of adolescent adjustment. Social experiences and maternal well-being (i.e., feelings of distress and psychological well-being) together accounted for 40.5% of the variance in adolescent adjustment. An additional 38.1% of the variance in adjustment was accounted for by the combination of speech difficulties, satisfaction with appearance, and use of social withdrawal as a method of cognitive, emotional, and social coping methods. Higher ratings of perceived speech difficulties, lower satisfaction with appearance, more frequent use of social withdrawal and distraction as coping strategies, and less frequent use of cognitive restructuring strategies were significantly related to poorer ratings of social experience, together accounting for over half (58.9%) the variance (Berger and Dalton, 2011). Edwards et al. (2011) showed that adolescents with CFA and more highly developed social skills were more likely to have made positive first impressions on others.

Studies that have examined ratings of self-satisfaction with appearance generally indicate a trend toward increased levels of satisfaction between early adolescence and young adulthood (Broder et al., 1994; Richman, 1997; Thomas et al., 1997), though further research is needed in order to better understand potential interactions between ratings of satisfaction, age, cleft type, and sex. For example, ratings of satisfaction with appearance were significantly higher in a group of 20-year-olds with cleft compared to younger CL/P cohort (e.g., 10- and 15-year-olds; Thomas et al., 1997). Results of at least one study by Broder et al. (1994) indicated the opposite trend for females with inter-oral defects such as iCP, in which ratings of satisfaction with appearance decreased rather than increased with age. Replication of these earlier studies on the adjustment of adults with craniofacial anomalies is needed.

Research by Turner et al. (1998) and Strauss et al. (1998) examining adolescent/young adult and parent satisfaction with cleft treatment and clinical outcomes has shown that perceived satisfaction with the clinical outcome of cleft treatment may differ between parents and

adolescents. Moreover, in the study by Turner et al. (1998), 23% of 15- and 20-year-olds with CL/P felt excluded from decisions regarding treatment planning. The results of these studies highlight the benefits of educating parents of older children and adolescents, who are growing in both independence and cognitive sophistication over the course of their development (Best and Miller, 2010), and about the importance of encouraging their children to become increasingly more involved in cleft-based care, treatment planning and medical decision-making. By taking a more active role in managing their medical care, adolescents and young adults should acquire additional knowledge or skills likely to better prepare them for later independent management of their condition; more readily identify barriers to effectively caring for and coping with CFA; provide opportunities in which to voice their preferences or concerns and make changes when problems arise; and improve overall satisfaction with the outcome of their cleft treatment.

Neuropsychological Aspects of Cleft

Early Development

Neuropsychological evaluation is limited in the early years of life (before 5 or 6 years of age) due to factors associated with both brain development (e.g., many cognitive skills have not yet been established and performance fluctuates over time yielding unreliable results) and personal characteristics of the child (e.g., difficulty in remaining on-task for the length of testing). Despite these measurement limitations, researchers have become adept at working with this young population and several consistent results in children with CL/P have been found.

One of the most well-documented findings in the literature on longitudinal outcomes of children with CL/P is increased risk for language deficits (Smith and McWilliams, 1968; Scherer and D'Antonio, 1995; Jocelyn et al., 1996; Neiman and Savage, 1997; Kapp-Simon and Krueckeberg, 2000; Morris and Ozanne, 2003). Toddlers (at age 5 to 36 months) with CL/P have exhibited significantly lower scores on measures of receptive and expressive language, vocabulary, and sentence complexity than non-cleft comparisons and normative data. At least one study by Schonweiler et al. (1999) has documented differences by cleft type and found poorer language outcomes for the CP group compared to the CLP group in toddlers at age 18 to 24 months. In contrast, Fox et al. (1978) found that language problems increased directly with cleft extensiveness. Millard and Richman (2001) also found differences in outcome by cleft type, with CP showing more educational problems than CLP and CL groups. Other studies have found no differences in language ability by cleft type (Broen et al., 1998). Furthermore, though it has been posited that the gap in language ability between children with CL/P and their non-cleft peers increases over time (Neiman and Savage, 1997; Smith and McWilliams, 1968), this finding has yet to be replicated (Kapp-Simon and Krueckeberg, 2000; Scherer and D'Antonio, 1995).

School Age

Cerebral maturation and associated increases in capacity for sustained attention/concentration and executive control and improved frustration tolerance theoretically permit a more thorough assessment of neurocognitive functioning in children with CL/P at school ages. Neuropsychological outcomes and subtle deficits in neurocognitive functioning in children with CL/P are of particular interest given this population's substantially increased risk for psychosocial, learning, and adjustment problems. Investigations of neurocognitive patterns by cleft type and gender have consistently found patterns of deficits in expressive language and verbal memory. One of the earliest neurocognitive studies by Lamb et al. (1973) found significantly lower verbal reasoning abilities (as measured by the Wechsler Intelligence Scale for Children [WISC] Verbal Comprehension Index [VCI]) in females in

the CLP group and males in the CP group compared with their female CP and male CLP counterparts. More recent comparison of Verbal and Performance Intelligence Quotients (VIQ and PIQ, respectively) in children with CLP, CP, and CL (Conrad et al., 2009) demonstrated an interaction of IQ and cleft type. Children with CLP had equal VIQ and PIQ scores (similar to healthy controls). Children with CP demonstrated the traditional discrepancy between average to above average PIQ but low average to below average VIQ. Finally, children with CL had high VIQ and low PIQ—a trend not previously documented. Comparisons between children with CL and CP have not been previously evaluated in this manner, and these findings provide potential evidence that cleft type may be associated with neurobiologically distinct conditions that are in turn associated with distinct neuropsychological, academic, and behavioral profiles. Such comparisons on verbal and perceptual skill between cleft types warrant further study and replication before conclusions can be drawn.

Patterns of language weakness have been studied extensively in this population (Richman, 1980; Richman and Eliason, 1984), and findings have indicated increased rates of specific, often subtle, weaknesses in language expression (i.e., verbal fluency), spontaneous verbal labeling of visual stimuli (i.e., rapid naming or automatic short-term memory), and rapid information processing. In a study by Richman (1980), patterns of language deficit varied by cleft type. When children with CL/P were divided into separate, diagnostically distinct language disorder categories, the verbal expressive disorders group (characterized by average receptive language abilities and below average expressive abilities) was predominantly comprised of children with CLP. The global language disorders group (characterized by below average receptive *and* expressive language abilities), in contrast, was predominantly comprised of children with CP. This group, characterized by a mixed receptive-expressive diagnostic profile, performed significantly below the verbal expressive group on measures of auditory short term memory, reading (decoding), and mathematics.

Recent research on the neurobiological aspects of cleft (Nopoulos et al., 2002c) has provided support for theories of abnormal brain structure in children with nonsyndromic clefts. Specifically, structural MRI data have shown that total brain size is decreased in children with CL/P. Within the smaller brain, there is specific reduction in volumes of the cerebellum, cerebral frontal lobe, and subcortical nuclei. Results also indicated gender differences in brain structure in cleft groups, with males exhibiting increased grey but decreased white matter volumes (Nopoulos et al., 2007). Furthermore, many of these abnormalities in brain structure have been directly correlated to cognitive, speech and behavioral deficits. For example, abnormal thickening of the ventral frontal cortex is directly correlated with inattention and hyperactivity (Nopoulos et al., 2010). Volumetric abnormality of the medial region of the frontal cortex was directly related to poor social function (Boes et al., 2007). Finally, decrements in the volume of the cerebellum were directly related to poor speech outcomes (Conrad et al., 2010). All of these studies have focused only on males and therefore the relationship between brain structure and function in females with CL/P requires further exploration.

Adolescence and Young Adulthood

Until recently, it was not well understood nor accepted that the neuropsychological deficits and related problems in academic, vocational, and social-emotional adjustment in individuals with CL/P persist despite the resolution of the physical cleft. The persistence of these difficulties over the lifespan, particularly with regard to speech and language functioning, has been consistently documented among individuals with CL/P (Pannbacker, 1975). Furthermore, studies have suggested that speech and language difficulties persist into adulthood at greater rates for individuals with CP than CLP or CL (Leeper et al., 1980). Studies of neuropsychological ability have also documented significantly lower

performances on tests of cognitive ability (e.g., Wechsler Adult Intelligence Scale, Third Edition [WAIS-III] VIQ and PIQ), verbal fluency, memory, and executive functioning in males with CL/P compared to non-cleft comparisons (Nopoulos et al., 2002b).

Consistent with the findings of neuroimaging with school-aged groups of children with CL/P, structural MRI data of men with CLP and CP documented global and regional volumetric differences. Specifically, adult males with CL/P exhibited normative intracranial, total brain, and cerebral volumes; but significantly decreased cerebellar size (which was attributed to global decreases in grey matter). Regionally, increases in cortical grey matter of the anterior cerebral volume, in conjunction with decreased posterior cerebral volume were observed. Abnormal volumes were associated with decreased global cognitive ability, as measured by WAIS-III full-scale IQ (Nopoulos et al., 2002a). The structural abnormalities observed in adult males with CL/P are distinct and suggest a potentially unique developmental pattern in cleft groups that warrants further examination.

Consistent evidence of subtle deficits in neuropsychological functioning is evident across all ages in individuals with CL/P, particularly with respect to speech and language abilities. These deficits are often related to cleft type, with CP groups and males in particular exhibiting the greatest impairment. Recent neurobiological data documenting structural brain differences between cleft and non-cleft groups provide compelling support for the potential influence of genetic/biological factors on cleft type, severity, and related longitudinal outcomes and for the role of brain structure abnormalities in neuropsychological dysfunction (Nopoulos et al., 2002a, 2002b, 2007).

Learning and Academic Aspects of Cleft

School-Age

Studies have generally indicated increased rates of academic problems in children with CL/P, even after controlling for IQ. Estimated rates of LD are much higher in CL/P groups, ranging from 30–40%, as compared to 10–20% in the general population (Broder et al., 1998). For example, in a recent study of caregiver ratings of psychosocial problems by age and cleft type, Snyder and Pope (2010) found that children with CL/P in the school age group (i.e., ages 4 to 11) exhibited three times the normative rates of problems in school competency. This age group showed higher rates of school problems than older adolescents with CL/P.

Though the presence of academic or school difficulties in children with CL/P is well-documented, the reasons for these difficulties are complex and not fully understood. However, careful analysis of the relevant literature highlights several variables (including expectancy effects and temperament) that have been shown to effect different outcomes at school ages. Richman (1978) found that the expectations of parents and teachers appear to have some influence on academic outcomes; for example, lower parent and teacher expectations for achievement have been associated with lower school functioning among children with CL/P. Richman (1978) also found that teachers tended to more frequently underestimate the intellectual ability of children with CP and more severe visible anomalies than they did for those with milder anomalies. Richman and Eliason (2008) documented an indirect relationship between behavioral inhibition and school achievement, such that academic performances were poorer in more inhibited children with CL/P.

Reading disabilities in children with cleft are highly prevalent and thus an area of particular emphasis within the CL/P literature (Richman, 1980; Richman and Eliason, 1984; Broder et al., 1998; Chapman, 2011; Collet et al., 2010b). Reading also represents a particularly salient aspect of cognitive and academic development in young children upon school entry.

Richman and colleagues have found elevated rates of reading disabilities in CL/P groups (36%), which are often thought to be associated with subtle neuropsychological deficits in language/verbal expression (i.e., deficits in verbal mediation and spontaneous verbal labeling of visual information), memory, and processing speed (Richman, 1980; Richman and Eliason, 1984; Richman et al., 1988; Richman et al., 2005).

Richman and Ryan (2003) found significantly lower rapid naming (1.5 standard deviations [SDs] below mean when compared to normative data) and verbal fluency (1 SD below the mean) in 8- to 11-year-old children with nonsyndromic CL/P and reading disability (n = 46) than in those with nonsyndromic CL/P without reading disabilities (n = 46) who were matched on the demographic variables of age, sex, and grade. No differences in phonemic awareness ability were found. Results of multiple regression analysis indicated that rapid naming, or automatic short-term memory for labels of visual stimuli, as assessed in this study by the Rapid Automatized Naming Test (Denkla and Rudel, 1976), was significantly and independently associated with reading level, suggesting that this aspect of expressive language may be of relatively greater importance to reading ability in children with cleft. Richman and Ryan's (2003) findings provided preliminary evidence that CL/P groups may not fit within traditional models of developmental Dyslexia, in which deficits in phonological awareness and memory have been emphasized as reliable predictors of reading level in children without CL/P. Based upon these results, the authors posited that a Dysnomia (or short term memory deficit) model of reading disability more adequately characterized individuals with CL/P (Richman and Ryan, 2003; Richman et al., 2005). This model has since facilitated recommendations for screening measures in children with CL/P in applied clinical settings (Yazdy et al., 2007).

Though increased risks for language deficits (i.e., verbal fluency and rapid naming) and associated academic problems (i.e., reading disabilities) have generally been well-documented in the area of language/reading in children with cleft (Collett et al., 2010b; Richman et al., 1988; Richman and Eliason, 1984), discrepancies in these findings have also been noted. For example, in a recent study comparing language and reading skills in 42 neurologically and genetically normal 5- to 7-year old children with CL/P to demographically similar children without cleft, Collett et al. (2010b) found significantly lower word identification and sound blending (as measured by Letter-Word Identification and Word Attack subtests of the Woodcock-Johnson Achievement Battery—Third Edition; WJ-III), phonological memory (as measured by Nonword Repetition and Digit Recall subtests of the Comprehensive Test of Phonological Processing; CTOPP), and word and nonsense word reading fluency (as measured by the Test of Word Reading Efficiency; TOWRE) in children with cleft relative to comparisons.

In contrast, a study by Collett et al. (2010a) that was published shortly thereafter failed to find statistically significant differences between CLP (n = 29) and CP (n = 28) groups and a demographically similar contrast group (n = 77) in vocabulary (as assessed by the Wechsler Preschool and Primary Scales of Intelligence—Revised; WPPSI-R, WISC-III; WISC-III, Expressive One-Word Picture Vocabulary Test—Revised, and Peabody Picture Vocabulary Test—Revised) or verbal fluency (as measured by the 'FAS' test). Furthermore, language functioning (as assessed by tests of vocabulary skill and verbal fluency) in both case and contrast groups was within the average range relative to test norms—with the exception of the fact that reading scores, as measured by a composite of Letter-Word Identification, Passage Comprehension, and Dictation subtests of the WJ-R, were significantly higher for children with cleft than those without (Collett et al., 2010a).

The discrepancies observed in these studies may be attributed to a number of factors, including: sample size/power-related differences; sample demographics such as age and sex;

cleft type and severity; aspects related to management of cleft palate-craniofacial care and research enrollment/participation; differences in rates of early identification of learning and behavioral problems and access to, for example, speech/language intervention or special educational services; and subtle but potentially important distinctions between dependent measures used to assess language and reading (i.e., vocabulary skill vs. verbal memory and rapid naming). The use of normative data for between-group comparisons without inclusion of a well-matched, demographically-similar contrast group of children without cleft has been described by Collett et al. (2010a) as a potential limitation to previous studies that might explain the failure in replicating previous findings (e.g., Richman et al., 1988; Richman and Eliason, 1984). The use of normative data, as Collett et al. (2010a) noted, may pose limitations to the extent that cleft and norm groups may differ demographically. This notion was not supported by their findings (i.e., cleft and contrast groups performed within the average range relative to norms and exhibited no significant differences in language performance), which failed to suggest significantly lower functioning in children with cleft regardless of method of comparison used.

Adolescence and Young Adulthood

In general, the research on academic/school outcomes among adolescents and young adults with CL/P suggests declining rates of LD, particularly reading disabilities, with age (from 48% at 6–7, to 22% at 8–9, to normal 8% rates by 10–13), though differences have been observed according to cleft type. Specifically, CP groups have shown higher rates of reading disabilities in younger children (53% at 6–7; Richman et al., 1988). While rates of reading disability (i.e., Dyslexia) have been found to be elevated in school-aged children with CL/P, gradual declines in rates of reading disability in children with CLP and CL have been observed when these same groups are studied into their adolescence and young adulthood. Rates of Dyslexia for CP groups, particularly when male-dominated, however, have failed to show similar rates of decline, remaining relatively higher (i.e., 33%) than the normative rates (9%) of their CLP and CL peers (Richman et al., 1988). The reason for this difference is unknown and should be further investigated. College attendance rates have been shown to be lower for children with CL/P compared to their siblings/non-cleft controls and lowest when rates were examined by cleft type, with the CP group showing the most discrepant college attendance rate (Danino et al., 2005; Marcusson et al., 2001).

Neuroimaging research has played a crucial role in our understanding of the relationships among behavior, subtle language deficits identified through neuropsychological testing, functional problems in learning or below average academic achievement, and brain structure/function. Through functional magnetic resonance imaging (fMRI), Nopoulos et al. (2007) found differences in regional blood flow between men with CL/P and non-cleft controls. Results further indicated that patterns of neural blood flow for men with nonsyndromic CL/P were comparable to the patterns of decreased blood flow through the neural circuit in conjunction with increased blood flow through the anterior circuit observed in individuals with diagnosed Dyslexia. Abnormal patterns of blood flow observed in men with CL/P during a reading task demonstrated increased blood flow to the right cerebellum, right occipital cortex, and left frontal operculum, and discussed as an indication of neural inefficiency (Goldsberry et al., 2006).

Summary

The current paper reviewed the empirical data on psychosocial/behavioral, neuropsychological, and learning/achievement functioning in children with CL/P across three age levels: infancy/early development, school age, and adolescence/young adulthood. The research highlights both the risks and protective factors associated with craniofacial conditions such as CL/P. The following is a summary of the key findings:

Psychosocial and Behavioral Outcomes

Psychosocial and behavioral outcomes during the early development of individuals with CLP are characterized by factors that have the potential to both increase parent emotional distress and foster increased attachment and bonding between parent and child. Levels of parent distress and quality of attachment are in turn associated with parent-child interaction problems and resiliency. School age outcomes are characterized by behavioral inhibition; externalizing behavior problems appear to vary by age and sex. Adolescent outcomes are characterized by increased concerns with speech and appearance and an associated decrease in self-confidence. Research shows parent-adolescent differences in perception of treatment outcome. Regardless of speech and appearance, participation in increased social activities and greater emphasis on and opportunity for involvement with peers during the adolescent years can be associated with both stress and resiliency. Studies have found that social experiences are highly correlated with psychological adjustment in adolescents with CL/P.

Neuropsychological Outcomes

Neuropsychological outcomes point to increased risk for language deficits, though some variability in the nature or severity of language deficits is observed by cleft type, particularly in studies of children of school age. Research on outcomes at school age indicates more subtle language problems (expressive language deficits such as Dysnomia). Adolescent outcomes indicate problems with verbal fluency, subtle memory problems, and cerebral volumetric differences between adolescents with and without CL/P.

Learning and Academic Outcomes

Research indicates a high incidence of reading problems and other LD among school-aged children with CL/P. It has been hypothesized that the increased risk for reading problems in children with CL/P is related to the subtle neuropsychological deficits (such as those observed in memory and rapid verbal labeling). Adolescence is characterized by declining rates of LD, though outcomes potentially differ by cleft type. Studies have shown lower college attendance among individuals with CL/P compared to that of siblings without CL/P. Functional neuroimaging studies have shown that patterns of abnormal neural blood flow in male adults with CL/P during a reading task are similar to patterns observed in non-cleft males with Dyslexia.

Recommendations

Primary prevention, skill-building, and targeted research-based interventions such as social skills training programs (e.g., Elman and Kennedy-Moore, 2003; Kapp-Simon et al., 2005) can play a significant role in minimizing the impact of risk on developmental outcomes in children and adolescents with CL/P. Research has shown that dissatisfaction with appearance (self-appraisal) may be an even better indicator of poor outcomes than objective variables (age, sex, cleft type). Children and adolescents with CL/P who have developed a realistic self-assessment, regardless of severity of speech or facial differences, are better adjusted than children with unrealistic self-assessment (Richman et al., 1985; Richman, 1991). It is also clear that more highly developed social skills play an important and protective role in minimizing stigma and enhancing the quality of social experiences in youth with craniofacial differences (Edwards et al., 2011). These findings strongly suggest the importance of prevention and intervention efforts which target adaptive social skill development; encourage realistic self-appraisal; and facilitate ongoing and open communication between parents and children about their condition.

The increased risk for learning problems in this population further highlights the importance of early identification and evaluation. Neuropsychological evaluation, including thorough

assessment of subtle language functions and pre-reading skills such as phonological awareness, rapid naming, and verbal fluency as well as memory and learning (reading), is often warranted for children with CL/P. The increased risks for language and reading problems should be discussed with parents by craniofacial teams, pediatricians or family practice physicians even if a child has passed earlier screening measures, as these instruments are often less sensitive to subtle problems such as those seen in children with CL/P.

Conceptual and Methodological Limitations of Prior Research

Variation in design and methodology contribute to variability in neuropsychological, behavioral, academic aspects of children with CL/P. Overall, research suggests that the outcomes addressed in the current paper can vary substantially depending upon age and developmental level. Research also suggests that findings in the area of orofacial clefting are a function of complex interactions among numerous variables including not only age but also gender and cleft type (Richman and Millard, 1997; Snyder and Pope, 2010). In general, studies examining psychosocial and behavioral outcomes in individuals with cleft indicate increased risk for internalizing and externalizing behavior problems in both males and females, though levels of behavioral and adjustment problems vary for males and females depending upon the age at which they are assessed (Richman and Millard, 1997). Therefore, a proportion of the seemingly contradictory findings in this area may instead represent demographically-driven differences that would be expected between studies employing samples that differ with regard to these characteristics.

Studies of CL/P outcomes have been limited by minimal use of combined parent, teacher, and self-report measures. Recent findings have highlighted key differences between physician and parent ratings of appearance and self-ratings of appearance (which are correlated with psychosocial outcomes) in addition to differences between parent and adolescent satisfaction with treatment outcome. Thus, findings based upon a single informant may provide limited and potentially biased information about a child's functioning. Small sample sizes and limited statistical power, which have likely contributed to some of the variability in outcomes across studies, have also limited analysis of potential interactions among sex, age, and cleft type. Demographic data have been reported and analyzed inconsistently.

The wide variety of instruments used to measure language, behavior, and learning has resulted in inconsistencies in outcome that are difficult to interpret in addition to limiting cross-study comparisons. The reliance upon global measures of cognitive functioning (e.g., WISC-IV) also poses a limitation as IQ data may underestimate the prevalence of subtle neuropsychological deficits in children with CL/P thereby limiting opportunities for identifying those who are at risk for problems in learning and achievement. Given that there may be associated conditions that are not yet well understood, it is also possible that some outcomes reviewed in the current paper apply only to a subset of the overall population. Until greater knowledge of associated genetic and neurodevelopmental factors is acquired, caution in generalizing from group data on cleft is advised.

Conclusion and Future Directions

Research on the psychosocial/behavioral, neuropsychological, and learning/academic outcomes in CL/P highlights that the findings are far from uniform and vary substantially by individual (genotypical and phenotypic) factors as well as by social, cultural, and environmental conditions. Further studies on the relationships among neuropsychological testing, functional problems in learning or below average academic achievement, and brain structure/function are warranted, particularly with school age (and younger) individuals with

CL/P and females. Further systematic research is necessary to better understand the impact of surgeries on psychological outcomes in individuals with cleft.

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