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Language and Early Reading Among Children With Orofacial Clefts

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Abstract

Objective—To test the hypothesis that children with orofacial clefts score lower than controls on measures of language and reading and to examine predictors of these outcomes.

Design—Longitudinal study tracking the development of children with and without orofacial clefts from infancy through age 7 years.

Subjects—Children with isolated cleft lip and palate (n = 29) and cleft palate only (n = 28) were recruited from the craniofacial program in an urban medical center. Seventy-seven demographically similar, unaffected controls were recruited via advertisements placed in area pediatric clinics and community centers.

Measures—Infant measures assessed child development and mother-child interactions during feeding and teaching tasks. At ages 5 and 7 years, measures of language functioning and academic achievement were completed along with an interview to collect school placement data and information on speech services received.

Results—There were no significant group differences in language at ages 5 and 7 years. Children with clefts scored significantly higher than controls on measures of early reading at age 7 years. Outcomes were predicted by demographic factors, the quality of mother-child interactions during teaching and feeding tasks, and cognitive development scores at age 24 months.

Conclusions—Findings do not support the hypothesis that children with clefts score lower than controls on neurocognitive and academic achievement measures. Predictive analyses revealed several dimensions that may be used in clinical practice to identify children at risk for learning and developmental concerns.

Keywords

language; longitudinal; orofacial cleft; reading

The results of case series studies (e.g., Kapp-Simon and Kruckeberg, 2000; Neiman and Savage, 1997) and a few case-control studies (e.g., Broen et al., 1998; Jocelyn et al., 1996;

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Speltz et al., 2000) indicate that infants and toddlers with cleft lip and palate (CLP) and cleft palate only (CP) score below average on clinician-administered assessments and parent-report measures of motor, cognitive, and language development. It is less clear that these early deficits portend later difficulties, although a growing number of studies of school-age children with clefts suggest that this is likely, depending on the skills assessed. Verbally mediated functions, such as verbal fluency, rapid naming, and auditory memory, are frequently reported weaknesses for children with clefts (Eliason and Richman, 1990). Recent work by Nopoulos et al. (2002) indicates that these verbal deficits continue to manifest in adulthood. Among individuals with different types of clefts, males with CP tend to score the lowest on cognitive measures (e.g., Richman, 1980). However, this finding has not been replicated consistently, with others showing that those with more severe clefts (e.g., bilateral CLP) have worse performance (Nopoulos et al., 2002).

In part, these mixed findings may be attributed to small sample sizes, sampling bias, limited statistical power, and the age of some of the earliest seminal studies (e.g., Richman, 1980; Richman et al., 1988). Furthermore, the risk of any type of neuropsychological deficit is elevated by the presence of associated malformations (Goodstein, 1961; McWilliams and Matthews, 1979; Swanenburg de Veye et al., 2002), and many studies of neuropsychological outcomes have not specified the nature of their samples in this regard.

Research on academic functioning has been more consistent and suggests that children with clefts are at risk for learning disabilities (LDs) and academic underachievement. For example, Broder and colleagues (1998) found that children with clefts showed higher than expected rates of LDs based on IQ versus achievement discrepancies, had higher rates of grade retention than observed in the general population, and scored below grade level on standardized tests. Boys with CP were at greatest risk, with roughly 80% meeting research criteria for a LD. Similarly, a recent population-based study by Yazdy et al. (2008) suggested that children with clefts were roughly twice as likely as noncleft peers to receive special education services. As might be expected, the most frequent service was speech-language therapy; however, rates of other special education services—including those for students with LDs, intellectual disabilities, and behavior disorders—were also higher among individuals with clefts.

Reading appears to be an area of vulnerability for children with clefts. For example, Richman et al. (1988) have shown that up to 53% of children with CP and 49% of children with CLP show evidence of a reading disability. Richman and Ryan (2003) have suggested that the nature of these reading problems may differ from those observed among noncleft children with dyslexia. Children with clefts, for example, may be more likely to show deficits in rapid naming rather than the phonological processing problems often seen in poor readers without clefts.

These findings may be anticipated by the association between embryological development of the face and brain, suggesting a possible biological vulnerability for children with craniofacial anomalies (Kjaer, 1995; Sperber, 1992). Research using psychophysiological measures and neuro-imaging technology suggests that differences in brain structure and functioning exist among individuals with either CLP or CP (e.g., Ceponiene et al., 2000; Nopoulos et al., 2001; Nopoulos et al., 2007). Environmental factors also may place children with clefts at risk. The quality of mother-child interactions has been the focus of several investigations (see review by Collett and Speltz, 2006). Findings from these studies have been inconsistent; however, there is some indication that the mothers of preschoolers with clefts are more directive than the mothers of unaffected controls, particularly during teaching interactions (e.g., Allen et al., 1990; Wasserman and Allen, 1985). For their part, children with clefts appear to be less assertive during conversational interactions with their

mothers than noncleft peers are with theirs (Frederickson et al., 2006). Allen et al. (1990) have hypothesized that the mothers of children with clefts may attempt to compensate for real or perceived delays by engaging in active teaching and direction. This style might ultimately be counterproductive, as suggested by studies of other medically fragile and typically developing children, indicating that maternal interactions characterized by a facilitative (rather than directive) style are conducive to cognitive and language development (e.g., Murray and Hornbacker, 1997).

Although the above findings are compelling, research in this area has been limited in at least two respects. First, with few exceptions, researchers have relied on test norms rather than demographically matched comparison or control samples. This method is problematic because cases drawn from clinic programs may differ substantially from the samples used to develop test norms (e.g., clinical samples may overrepresent or underrepresent urban or rural families or families with particular socioeconomic backgrounds). Test norms also may become invalid over time, due to changes in population characteristics and/or environmental factors (Flynn, 1984, 1987). As a result, the deficits suggested by previous studies may be better explained by demographic factors (socioeconomic status, gender, or ethnicity) than by participants' cleft status. Second, there are few longitudinal investigations documenting the emergence of cognitive and early academic skills and their development over time. Clinically, the identification of factors that best predict later neuropsychological development has tremendous value because children with clefts at the highest risk for adverse outcomes could be targeted for early intervention and preventative efforts. Further study of these issues is timely because the American Cleft Palate–Craniofacial Association's (ACPA) recently published standards for team care advocate for routine assessment of cognitive development and learning among children with craniofacial anomalies (ACPA, 2008). Data on the nature and extent of neurocognitive differences between cases and controls are needed to guide such evaluations.

The purposes of this study are twofold. First, we sought to test the hypothesis that children with clefts would score lower than demographically matched controls on language measures at ages 5 and 7 years and on measures of basic reading at age 7 years. Second, we sought to explore the value of demographic, medical, and psychosocial factors in infancy for predicting later cognitive and academic outcomes. Participants were a cohort of children followed from ages 3 months to 7 years, giving us the opportunity to determine which factors, commonly assessed in clinical practice, would help to identify those children at greatest risk for cognitive deficits and academic failure.

Subjects and Methods

Participants

CLP and CP—Children with clefts were drawn from consecutive referrals to the craniofacial center at an urban children's hospital. The families of children with clefts were approached for participation at the time of diagnosis by one of the pediatricians from the craniofacial team. Children with clefts were included if they had a diagnosis of CLP (n = 29) or CP (n = 28). Infants with additional birth defects, identified genetic syndromes (e.g., 22q11 deletion), or perinatal problems known to affect cognitive development (e.g., preterm birth) were excluded.

Controls—Control group infants (n = 77) were recruited via advertisements placed in local pediatric clinics and community centers. Infants were included in this group if they matched a CLP or CP case on the basis of socioeconomic status (SES), gender, and age. Twenty of the 77 control group participants were demographic matches for children in another craniofacial comparison group (sagittal synostosis) who were part of our original study but

were not included in the current analyses. Because the 20 matching control group participants met all inclusion and exclusion criteria and were treated identically to other participants, they were included in the current analyses in order to maximize sample size and statistical power. In addition to the exclusionary criteria listed for children with clefts, control participants were excluded if they had a history of any craniofacial anomaly.

Measures

Infant Development and Medical/Developmental History—Infants were assessed using the Mental Development Index and Psychomotor Development Index (MDI and PDI, respectively) from the Bayley Scales of Infant Development (BSID; Bayley, 1969) when they were 12 and 24 months old. Medical history and demographic data were collected using parent interviews and medical records reviews, with updates collected at each time point.

Mother-Infant Interaction—The quality of mother-child interactions was assessed at ages 3, 12, and 24 months using the Nursing Child Assessment of Feeding Scale (NCAFS) and Nursing Child Assessment of Teaching Scale (NCATS; Barnard et al., 1989). The NCAFS and NCATS are observational scales that assess both parent and child behaviors. The NCAFS is administered during feedings, with parents instructed to feed the child as they would at home. The NCAFS includes 76 items scored on a dichotomous yes/no scale to indicate whether target behaviors were observed during the feeding. Scores can be derived separately for parent and child domains and for a composite or total score reflecting both parent and child behavior. Within the parent domain, subscales include sensitivity to cues, response to distress, social-emotional growth fostering, and cognitive growth fostering. Child domain scores include clarity of cues and responsiveness to caregiver. The NCATS is comparable in format and is administered during parent-child teaching interactions. The NCATS includes 73 items scored on a dichotomous yes/no scale. Scores are derived for parent and child skills in the same domains as the NCAFS, as well as a total score. The NCAFS is intended for infants from birth to 12 months, and the NCATS can be completed with infants and young children from birth to 36 months. Reliability and validity data are provided by Barnard et al. (1989). For this study, the NCAFS was completed when children were ages 3 and 12 months, and the NCATS was completed at ages 12 and 24 months. Total scores from the NCAFS and NCATS were used for analyses. Interactions were videotaped and coded by trained observers, with one coder rating the majority of participants and three additional coders providing reliability checks on a randomly selected subgroup of roughly 50%. Interrater agreement was 91.2% across all items for the NCAFS (mean κ = .65) and 86.2% for the NCATS (mean κ = .59).

Language Skills—Expressive and receptive language skills were assessed using a battery of standardized assessments, including vocabulary subtests from the Wechsler Preschool and Primary Scales of Intelligence–Revised (Wechsler, 1989; age 5 years) or the Wechsler Intelligence Scale for Children–III (Wechsler, 1991; age 7 years); the Expressive One-Word Picture Vocabulary Test–Revised (Gardner, 1990); the Peabody Picture Vocabulary Test–Revised (Dunn and Dunn, 1981); and a verbal fluency test (Benton, Hamsher, and Sivan, 1994). In the interest of parsimony, and because these measures were moderately to highly correlated (i.e., r = .46 to .73 at age 5 and .42 to .76 at age 7), we chose to derive a composite language score by converting test scores to standard scores (100 [15]) and calculating an average.

Reading Achievement—At age 7, the letter-word identification, passage comprehension, and dictation subtests from the Woodcock-Johnson–Revised (WJ-R; Woodcock and Johnson, 1989) were administered to assess early reading and related skills. Letter-word identification is a measure of letter knowledge and single word reading that requires the

child to read a series of increasingly difficult words. Passage comprehension is a measure of reading comprehension, requiring the child to read a sentence and fill in a missing word. Dictation measures spelling ability, a skill closely related to children's phonemic decoding. Again, these variables were highly correlated (r = .76 to .90), and a composite standard score reflecting the average of these subtests was derived.

Special Education and Speech Services—Parents completed a brief interview when children reached age 7, which included questions about grade placement and special education services received. Children were categorized as receiving special education if they had received or were currently receiving services. Parents also reported on their child's special education classification using categories that corresponded to the Individuals with Disabilities Education Act (communication disordered, specific learning disability, severe behavior disorder, health impaired, visual impairment, orthopedically impaired, or other). Additional information was collected regarding parents' concerns about their child's speech and services received (no concerns; speech concerns discussed with a health care provider, but not formally evaluated; speech concerns evaluated, no treatment planned; speech concerns evaluated, treatment planned; speech concerns evaluated, treatment in progress or completed).

Procedures

Study visits were scheduled at ages 3, 12, and 24 months, with follow-ups completed at ages 5 and 7 years. All assessments were completed within a month of the child's target age. Tests were administered by trained examiners, in accordance with standardized instructions. Child testing and mother-child interactions took place in a clinic room with an observation window that concealed videotaping equipment. A variety of strategies were used to facilitate retention over the course of the study, including holiday greeting cards for families and birthday cards for children, monetary incentives, and feedback letters summarizing the results of child evaluations. Parents provided informed consent prior to participating, and children provided assent at ages 5 and 7 years. First wave data collection (i.e., assessments at age 3 months) began in 1989, and the final age 7 data were collected in 2000.

Statistical Analysis

Descriptive analyses (e.g., frequencies, means, and standard deviations) were used to compare the demographic background and prenatal and birth histories of the cleft and control groups. Analyses used comparisons of each cleft group with the pooled control group, rather than paired comparisons of matched cleft cases and controls, in order to take advantage of the gain in precision from the use of the larger control group. As noted, composite scores were developed by domain to reduce the number of group comparisons. Regression analyses were used to compare the cleft groups with controls while adjusting for demographic variables (e.g., gender, race/ethnicity, SES). Given the evidence of differential attrition by SES, we also examined these data using inverse probability weighting (IPW; Heyting et al., 1992). IPW accounts for differential attrition using an estimated probability of continued participation in the study for each participant. In essence, this approach provides additional "weight" for participants who were retained in the study but were likely to have been lost to follow-up based on demographic or other characteristics. As used here, we predicted attrition using demographic variables (SES, parents' marital status, sex, ethnicity), 24-month MDI scores, 12-month NCAFS scores, and composite language scores from age 5. We then generated scores reflecting the probability of each participant being observed at age 7 years. Regression analyses were then rerun, weighted by the inverse of the probabilities of being observed, to determine whether there was a meaningful change in results. Odds ratios, adjusted for demographic variables as described above, were used to compare the rates of special education and speech services for children with and without

clefts. Predictive analyses, investigating the variables that best predicted language and reading outcomes at ages 5 and 7, were conducted using stepwise multiple regression. This study was approved by the Seattle Children's Hospital Institutional Review Board.

Results

Attrition

Data were available for 86% of the sample at age 5 (50/57 cases, 65/77 controls) and 70% at age 7 (41/57 cases, 53/77 controls). There was some evidence of differential attrition as a function of socioeconomic status, with lower SES families more likely than higher SES families to drop out. For example, at age 5 years, 91% of the families in SES categories 1 through 3 were retained; whereas, only 81% of those in SES categories 4 and 5 were retained. At age 7 years, 80% and 46% of the families in SES categories 1 through 3 versus 4 and 5 were retained, respectively.

Demographics

The demographic characteristics of the sample are summarized in Table 1. Consistent with the base rates for orofacial clefts, the majority of the children with CLP were boys (64%); whereas, the majority of the children in the CP group were girls (75%). The majority of children were from middle class households (i.e., Hollingshead SES categories 2 through 4; Hollingshead, 1975). Most of the children were non-Hispanic white (84%), which reflects the ethnic distribution of the patient population served.

Birth and Parental Factors

Descriptive analyses showed that children with clefts were more likely to be premature and less likely to be delivered by cesarean section compared with control children (Table 2). Children with CLP tended to have slightly lower birth weights and longer hospital stays than others. Not surprisingly, control children were much more likely to be breastfed as infants than were children with clefts. According to parent report, children with clefts were less likely to begin speaking by age 12 months than were control children. Scores on the NCAFS and NCATS were slightly lower for children with clefts than for control children.

Language and Early Academic Outcomes

As seen in Table 3, all groups scored within the average range on language measures relative to test norms. Case-control differences were small in magnitude (i.e., adjusted differences of approximately 3 to 5 standard score points) and were not statistically significant (Table 4). In reading, patients scored higher than unaffected controls ($p = .03$). This difference was most apparent for those with CP, with an adjusted difference of just over 10 standard score points. As seen in Table 5, analyses using IPW did not result in significant changes in the findings for language or reading outcomes. This suggests that any bias due to differential attrition was modest in relation to these outcomes and is unlikely to account for the findings.

Special Education and Speech Services

Based on parent report, rates of special education placement were 19.3% for controls, 56.5% for children with CLP, and 40.9% for children with CP (Table 6). Odds ratios adjusted for demographic factors suggest that children with CLP were significantly more likely than controls to receive special education services (adjusted odds ratio = 3.8, $p = .02$). Though not statistically significant, a history of special education placement was also more likely for those with CP (adjusted odds ratio = 3.6, $p = .07$). The most frequently reported special education classification for children with and without clefts was communication disability,

followed by specific learning disability (Table 6). Speech concerns were reported more frequently by the parents of children with CLP (43.5%) and CP (36.4%) compared with the parents of controls (22.8%). As seen in Table 7, speech had been formally evaluated in most children with CLP (65.3%), many with CP (36.5%), and relatively few controls (14.1%). Children with CLP were much more likely than controls to have received speech therapy, after adjusting for demographic characteristics (adjusted odds ratio = 12.1; $p = .001$). Children with CP were less likely than controls to have received speech therapy, though this difference was not statistically significant (adjusted odds ratio = 5.0; $p = .14$).

Prediction of Outcomes: All Children

The predictive value of demographic factors varied widely across outcome variables. Demographics accounted for 38% to 41% of the variance in age 5 and age 7 language scores, and 20% of the variance in reading scores (Table 8). At age 5, NCAFS scores (F_{Δ} 2, 54 = 4.6, $p = .01$) and BSID scores (F_{Δ} 2, 52 = 16.5, $p < .001$) added significant predictive value. At age 7, NCAFS scores (F_{Δ} 2, 49 = 5.5, $p = .007$), BSID scores (F_{Δ} 2, 47 = 23.7, $p < .001$), and age 5 language scores (F_{Δ} 1,46 = 21.1, $p < .001$) all added predictive value for language outcomes. NCATS scores contributed significantly to the prediction of reading outcomes (F_{Δ} 2,48 = 6.2, $p = .004$).

After adjusting for demographic and other variables, BSID MDI scores ($\beta = .56$, $p < .001$) and NCAFS scores ($\beta = .15$, $p = .04$) remained significant predictors of language outcomes at age 5. As expected, the composite language score at age 7 was related strongly to the age 5 language score ($\beta = .50$, $p < .001$). Even after controlling for age 5 scores, the BSID MDI score at 24 months was predictive of age 7 scores ($\beta = .35$, $p = .002$). Composite reading scores at age 7 were predicted by use of spoken words by age 12 months ($\beta = .27$, $p = .04$), NCATS scores ($\beta = .27$, $p = .04$), and cleft diagnosis ($\beta = .33$, $p = .02$) after controlling for age 5 language scores.

Discussion

This study compared the reading and language skills of children with orofacial clefts with those of a matched control group. It is the first to follow a sample of children with clefts from infancy to the elementary school years. Recruitment of the sample, therefore, occurred at a time well before the possibility of known or suspected academic problems, reducing the potential for sampling bias (e.g., families participating because they were concerned about their child's academic progress). The longitudinal design of this research also provided the opportunity to examine early-life predictors of subsequent performance on measures of language and basic reading skills.

After adjusting for potential confounds, we found small and statistically nonsignificant differences in language between children with clefts and those without. Patients and controls both scored roughly within the average range relative to test norms, and differences by cleft type were minimal. To our surprise, children with CP scored higher than controls in early reading, and the magnitude of this difference was relatively large (i.e., 10 standard score points).

These results, particularly the finding of higher reading scores among patients and equivalent functioning of children with CP and CLP, are contrary to expectations set by two previous studies. Richman and Eliason (1984) found that children with CP (ages 8 to 13) scored much lower than well-matched children with CLP on measures theoretically associated with reading skill, including auditory memory and verbal fluency. The children in this study's CP group were extremely low functioning, with group averages on some measures nearly two standard deviations below the normative mean. In a later study,

Richman et al. (1988) examined reading proficiency in children ages 6 to 13 with orofacial clefts. Although children with CP and CLP performed about the same on a measure of word recognition, children with CP had poorer reading comprehension than those with CLP, a difference that became more pronounced with age. The incidence of "reading disability" in the sample also was calculated (defined as a score at least one standard deviation below the mean of the test standardization sample). With increasing age, the differences between children with CP and those with CLP became more apparent; by ages 10 to 13 years, fewer than 10% of children with CLP had a reading disability; whereas, nearly a third of those with CP were considered reading disabled.

Our failure to replicate these findings may reflect differences in the severity of clefting or in other clinical features known to be associated with development (e.g., number of associated physical anomalies). Although both the current study and Richman's research sought to exclude children with known genetic syndromes, neither study gathered (or reported) detailed information about cleft severity or the number of associated minor physical anomalies (MFA). Future studies comparing CP and CLP groups would benefit from detailed sample characterization in this regard (e.g., reporting on the number and type of MFAs). Alternatively, children in this study may have been a lower risk group by virtue of their demographic background. Unfortunately, demographic data from the early studies by Richman et al. are limited, making it difficult to know how our samples compare. A third possibility, and perhaps the most compelling, is related to the speech and language therapy services received by children with clefts in our sample. Approximately one third to one half of the children with orofacial clefts in our sample had received speech intervention. It is possible that such intervention attenuated deficits in reading-related skills such as phonological processing and may have reduced disparities in basic reading. Comparable data are not provided in earlier studies by Richman and colleagues, making it impossible to determine how the services received by children in our sample might differ from those in previous studies. Overall, recruitment from a single site, even one serving a large and diverse patient population, is problematic. Language and reading are both related to demographic characteristics and may be associated with other clinical characteristics in orofacial clefting. Furthermore, screening and referral practices may differ substantially among different sites, as may the nature of services available in the community.

Despite the near equivalence of language scores in children with clefts versus controls and the slight advantage for the former in reading, children with clefts (particularly those with CLP) were more likely than controls to have a history of special education service, most often under a classification of "communication disability." Moreover, the parents of children with clefts frequently expressed concerns about their child's speech, and children with clefts were more likely to have received formal speech evaluations and speech therapy. Given the discrepancy in our data between tested performance and special education classification and speech therapy services, it seems likely that most children were being served for articulation problems, which were not formally assessed in this project (versus other language and learning problems). Although this finding is commensurate with the study by Yazdy et al. (2008), rates of special education service were higher for all groups in our study. This likely reflects regional differences in criteria for special education classification.

From a clinical perspective, our findings regarding the early predictors of outcome are more important than those pertaining to group differences. Demographic factors, mother-child interaction observations, and developmental assessments conducted when children were 24 months old proved to be robust predictors of later language functioning and reading achievement for both patients and controls. These findings highlight the need for future studies with broader demographic representation to elucidate the risk(s) conferred by having a cleft in addition to other known risks. Our finding regarding the importance of early

mother-child feeding and teaching interactions is consistent with a large body of research on the importance of maternal responsiveness in the development of cognitive and early academic skills, particularly among at-risk children (e.g., McGrath et al., 1998; Murray and Hornbacker, 1997). In contrast to previous studies of the power of infancy measures to predict school-age IQ (see Colombo, 1993, for a review), scores on the mental scale of the BSID at age 24 months were good predictors of language outcomes in this study. This may reflect the heavy language emphasis of the BSID for 24-month-olds and our prediction of language-specific measures, rather than global IQ.

These findings point to factors that may be used to prioritize screening of high-risk children with orofacial clefts in clinical practice. With their frequent craniofacial clinic visits, children with clefts are a “captive audience.” Routine screening for LDs and/or factors that dispose children to learning problems (as advocated by ACPA’s recent team care guidelines) may help to identify children at risk and steer them toward needed intervention. Demographics are an obvious and cost-effective starting point, which we found to account for a substantial portion of the variability in language and reading outcomes (i.e., demographic characteristics alone accounted for 20% to 41% of the variance in language and reading scores). Children from lower SES backgrounds appear to be at greatest risk and, by virtue of their SES, may have the most difficulty accessing early intervention services in the community. Increased screening efforts, using early childhood measures like the BSID and observational measures like the NCATS, may therefore be directed toward this population.

Several limitations of this study and directions for future research should be noted. Our measures of language function focused on vocabulary and there are other relevant dimensions of language that were not assessed, such as syntax, morphology, and verbal memory. Furthermore, we did not have an objective measure of articulation, which is an area of weakness for many children with orofacial clefts into the early school years (e.g., Chapman et al., 1998; Peterson-Falzone, 1995). Indeed, parent-report data suggest that this was an ongoing concern for children in our sample, particularly those with CLP, as indicated by their frequent special education placement and receipt of speech therapy. Although our basic reading measures were comparable to those used in other studies (e.g., Richman et al., 1988) and in many school systems, these instruments may not have been sensitive enough to detect differences in early reading among young children. Indeed, our study did not assess several important precursors of reading, such as phonological processing and rapid naming. Richman and colleagues (e.g., Richman and Ryan, 2003; Richman et al., 2005) have found rapid naming to be a particularly important determinant of reading for children with clefts. This is an important issue that warrants further study using state-of-the-art reading batteries. For young school-age children, such batteries should include phonological awareness, letter knowledge, single-word reading, nonword reading, and phonological memory (e.g., nonword repetition). For older children, who have typically mastered single word reading and begin reading for meaning, measures also should include those assessing fluency, comprehension, and written expression. Finally, our assessment of special education and speech services received relied on parent report. Ideally, we also would have been able to access educational and other service records to verify these reports and to determine the specific nature and intensity of services received (e.g., focus of intervention, number and duration of sessions).

Despite significant efforts to engage and retain families in the study, attrition at age 7 years was problematic. Although group differences did not appear to be influenced by attrition bias, this limited our statistical power to detect differences at age 7. Furthermore, this makes it difficult to determine whether regression models predicting outcome are applicable across demographic subgroups. The attrition in the later years of this study reflects the difficulties

associated with retaining high-risk families in research. Such families are likely to move more often, and, of necessity, research participation becomes a low priority. Alternatively, it may be that families were motivated to continue their participation in this research prior to their child's school entry (i.e., age 5) but became less engaged upon learning that he/she was functioning well. Longitudinal research should nonetheless remain a high priority as the best way to evaluate developmental trends in cleft populations. In addition, the results of this study highlight the importance of tracking those children who are the most difficult to track (i.e., with single parents who work long hours, from lower SES families). Finally, this study did not have adequate statistical power to detect relatively subtle differences in reading and academic functioning or to conduct extensive subgroup comparisons (e.g., by gender or SES). Large-scale, multi-site studies are needed to achieve those aims. Such studies will have the potential to address issues of sampling variability, helping to clarify whether differences in referral, screening, and intervention (e.g., for speech and language or special education services) account for disparate findings.

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TABLE 1

Comparison of Groups on Demographic Variables

Variable	Cleft Lip and Palate	Cleft Palate Only	Comparison
Gender *			
Male	20 (70%)	7 (25%)	43 (57%)
Female	9 (30%)	21 (75%)	32 (43%)
Child ethnicity *			
White non-Hispanic	26 (88%)	23 (82%)	62 (84%)
Hispanic	1 (4%)	0	0
African American	1 (4%)	0	5 (6%)
Asian	0	3 (11%)	1 (1%)
Other	1 (4%)	2 (7%)	7 (9%)
First born? *			
Yes	13 (40%)	10 (35%)	31 (41%)
No	16 (60%)	18 (65%)	44 (59%)
Number of siblings *			
0	14 (46%)	10 (35%)	32 (42%)
1	9 (31%)	8 (28%)	23 (30%)
2	4 (15%)	6 (21%)	15 (20%)
3 or more	2 (7%)	4 (14%)	5 (6%)
Hollingshead SES category *			
1 (high)	1 (3%)	5 (18%)	4 (5%)
2	6 (21%)	4 (14%)	29 (38%)
3	10 (34%)	9 (32%)	13 (18%)
4	4 (15%)	6 (22%)	16 (22%)
5 (low)	8 (28%)	4 (14%)	13 (18%)
Marital status *			
Single	3 (10%)	4 (14%)	14 (18%)
Married	26 (90%)	24 (86%)	61 (82%)
Mother's work status			
Works outside home	9 (32%)	7 (25%)	14 (19%)
Does not work outside home	19 (68%)	21 (75%)	61 (81%)
Mother's age †	26 (6)	27 (6)	29 (6)
Father's age †	27 (6)	30 (5)	32 (7)

* Figures are presented as number of subjects in each category (percentages within diagnosis groups); SES = socioeconomic status.

† Figures are presented as mean (standard deviation).

TABLE 2

Comparison of Groups on Birth Outcomes, Health, and Parental Factors From Birth to 2 Years of Age

Variable	Cleft Lip and Palate	Cleft Palate Only	Comparison
Type of delivery *			
Cesarean section	3 (12%)	5 (21%)	16 (24%)
Forceps	1 (4%)	0	3 (4%)
Natural	22 (85%)	19 (79%)	49 (72%)
Premature *			
Yes	11 (39%)	3 (12%)	11 (15%)
No	17 (61%)	22 (88%)	54 (85%)
Length of baby's hospital stay *			
1 day or less	3 (11%)	5 (19%)	16 (22%)
2 days	9 (33%)	9 (33%)	24 (33%)
3 days	5 (19%)	8 (30%)	15 (21%)
4 or more days	10 (37%)	5 (19%)	17 (24%)
Birth weight (lb) †	7 (1)	7 (1)	7 (1)
Mode of feeding (3 months) *			
Breast	2 (7%)	3 (11%)	50 (67%)
Other	27 (93%)	25 (89%)	25 (33%)
Age child began speaking *			
12 months or earlier	13 (44%)	12 (55%)	41 (69%)
Later than 12 months	14 (56%)	10 (45%)	18 (31%)
NCAFS †			
3 months	54 (10)	56 (8)	60 (7)
12 months	62 (5)	61 (6)	63 (5)
NCATS †			
12 months	56 (6)	57 (5)	58 (6)
24 months	60 (6)	61 (5)	62 (5)

* Figures are presented as number of subjects within each category (percentages within diagnosis groups).

† Figures are presented as group means (standard deviations); NCAFS = Nursing Child Assessment of Feeding Scale; NCATS = Nursing Child Assessment of Teaching Scale.

TABLE 3

Group Means (and Standard Deviations) of Neuropsychological Outcomes by Diagnosis Group

Variable	Cleft Lip and Palate	Cleft Palate Only	Comparison
Language functioning–5 years			
WPPSI-R vocabulary *	10 (4)	10 (4)	11 (3)
EOWPVT-R †	93 (20)	96 (22)	99 (21)
PPVT ‡	96 (20)	98 (19)	99 (22)
Verbal fluency §	12 (4)	12 (5)	13 (6)
Composite score	96 (15)	98 (18)	101 (16)
Language functioning–7 years			
WISC-III vocabulary ‖	11 (4)	12 (4)	12 (4)
EOWPVT-R †	104 (24)	104 (20)	111 (21)
PPVT ‡	99 (17)	99 (19)	108 (15)
Verbal fluency §	21 (6)	20 (6)	22 (6)
Composite score	101 (15)	102 (15)	108 (15)
Reading–7 years			
WJ-R LW ¶	107 (17)	110 (17)	100 (20)
WJ-R PC #	107 (16)	108 (17)	101 (18)
WJ-R Dict **	95 (12)	102 (11)	93 (16)
Composite score	103 (14)	107 (14)	98 (17)

* Vocabulary subtest, Wechsler Preschool and Primary Scales of Intelligence–Revised.

† Expressive One-Word Picture Vocabulary Test.

‡ Peabody Picture Vocabulary Test.

§ Benton Verbal Fluency Test.

‖ Vocabulary subtest, Wechsler Intelligence Scale for Children–III.

¶ Letter-word identification, Woodcock-Johnson Achievement Battery–Revised.

Passage completion, Woodcock-Johnson Achievement Battery–Revised.

** Dictation, Woodcock-Johnson Achievement Battery–Revised.

TABLE 4

Estimated Group Effects (± Standard Errors) From Regression Analyses Controlling Demographic Factors (See Model II in Table 8)

Variable	Cleft Lip and Palate Versus Control	Cleft Palate Only Versus Control	p Value for Group
Language composite–5 years	−3.41 ± 3.65	−2.52 ± 3.88	.53
Language composite–7 years	−5.24 ± 3.77	−5.34 ± 3.87	.19
Reading composite–7 years	5.74 ± 4.65	10.27 ± 4.73 *	.03

* $p = .03$.

TABLE 5

Estimated Group Effects (± Standard Errors) From Regression Analyses at Age 7 Years Using Inverse Probability Weighting

Variable	Cleft Lip and Palate Versus Control	Cleft Palate Only Versus Control	p Value for Group
Language composite–7 years	−2.14 ± 4.17	−3.57 ± 3.60	.31
Reading composite–7 years	6.52 ± 5.06	10.10 ± 4.47 *	.02

* $p = .03$.

TABLE 6

Frequency of Special Education Placement for Children With and Without Clefts

Special Education Category	Cleft Lip and Palate, n (%)	Cleft Palate Only, n (%)	Comparison, n (%)
Any category	13 (57)	9 (41)	11 (19)
Communication disability	11 (85)	4 (44)	4 (36)
Specific learning disability	1 (8)	3 (33)	3 (27)
Severe behavioral disorder	0	0	1 (9)
Other health impairment	1 (8)	0	1 (9)
Visual impairment	0	0	0
Orthopedic impairment	0	0	0
Other	0	0	0
Unknown	0	2 (22)	2 (18)

TABLE 7

Frequency of Speech Concerns and Services for Children With and Without Clefts

Speech Concerns and Services	Cleft Lip and Palate, n (%)	Cleft Palate Only, n (%)	Controls, n (%)
None	8 (35)	10 (46)	44 (77)
Speech concerns discussed with health care providers, not formally evaluated	0	0	1 (2)
Speech evaluated, no treatment planned	4 (17)	6 (27)	2 (4)
Speech evaluated, treatment planned	1 (4)	1 (5)	1 (2)
Speech evaluated, treatment in progress or completed	10 (44)	1 (5)	5 (9)

TABLE 8

Percentage of Variance Explained (R^2) From Regression Analyses of Age 5 and Age 7 Language Outcomes and Age 7 Reading Scores

Model	Predictors	Language (5 Years)	Language (7 Years)	Reading (7 Years)
I	Demographic factors *	.41	.38	.20
II	Model I and cleft diagnosis †	.41	.40	.23
III	Model II and child factors from birth to 12 months ‡	.41	.45	.31
IV	Model III and parent-child interaction at 12 months §	.54	.56	.44
V	Model IV and Bayley Scales of Infant Development (BSID) scores at 24 months ‖	.72	.79	.50
VI	Model V and language at age 5 ¶	—	.86	.52

* Gender, race/ethnicity, socioeconomic status, age at assessment, mother's marital status, mother's work hours, mother's age, and number of siblings.

† Three groups: cleft lip and palate, cleft palate only, control.

‡ Mode of delivery, birth weight, breastfed, began speaking by 12 months.

§ NCATS: Feeding, NCATS: Teaching; NCATS = Nursing Child Assessment of Teaching Scale.

‖ BSID MDI and PDI scales; MDI = Mental Development Index; PDI= Psychomotor Development Index.

¶ Composite language scores.