Objective   To examine reading and related skills in children with and without orofacial clefts.

Methods   Forty-two children with orofacial clefts were recruited from an urban craniofacial center. A
demographically similar sample of 43 children without clefts was recruited using community advertisements
and a research registry. Participants completed assessments of basic reading, phonological awareness,
phonological memory, reading fluency, and rapid naming. Parents completed a semi-structured interview
regarding educational and medical history.  Results   Children with clefts scored significantly lower than
controls on measures of basic reading, phonological memory, and reading fluency.  Conclusions   This is
one of the first studies of reading in children with orofacial clefts to include a control sample. The findings
suggest that children with clefts are less adept readers than demographically matched peers without clefts,
supporting the need to monitor academic achievement in this population.

Key words   academic achievement; cleft palate; orofacial cleft; reading.

There are many reasons to suspect that children with isolated (i.e., nonsyndromic) orofacial clefts may have
sharply elevated risk for reading and related learning problems. Citing the adage “the face predicts the brain”,
researchers have long hypothesized neurobiological differences between children with and without clefts, based on
the close connections between the embryonic development of facial and brain structures (e.g., Ceponiene et al., 2000;
Nopoulos et al., 2001). In support of this position, Nopoulos and colleagues (2001) found a higher incidence
of midline brain defects among adult males with clefts who were given structural MRIs. In more recent work, these
researchers reported volumetric brain differences between cases and controls in both child and adult samples
(Nopoulos et al., 2002; Nopoulos, Langbehn, Canady, Magnotta, & Richman, 2007). Though functional imaging
studies are limited, in one study Goldsberry, O’Leary, Hichwa, & Nopoulos (2006) found that individuals with
clefts exhibited neural inefficiency during reading and language tasks that was comparable with that observed
in adults with dyslexia. Measures of evoked response potentials (ERPs) have suggested that infants with clefts
respond differently than typical infants to auditory stimuli (Ceponiene et al., 2000). Alternatively, it may be the
sequelae of clefting that place children at-risk. Children with cleft lip and palate (CLP) and cleft palate only (CP),
in particular, suffer from early speech deficits due to the architecture of their lip and palate. Although this improves
following surgical repair in infancy and early childhood (Jones, Chapman, & Hardin-Jones, 2003), persistent
speech problems are observed among many children with clefts into school-age (Chapman, Graham, Gooch,
& Visconti, 1998; Peterson-Falzone, 1995). This may complicate their acquisition of phonological processing
skills and early reading ability. Similarly, otitis media is nearly ubiquitous among children with CLP and CP,
and may result in hearing deficits that complicate language acquisition and the development of phonological
skills.

Indeed, multiple studies have revealed that children with clefts are more likely to demonstrate learning
problems and to score below average on reading measures relative to test norms. Broder, Richman, and Matheson
(1998) reviewed medical charts and educational records
and found that children with clefts had elevated risk for learning disabilities (defined as a discrepancy between IQ and scores on group achievement measures such as the Iowa Tests of Basic Skills). Forty-six percent of the children with clefts in their sample showed evidence of a learning disability, with rates highest among males with CP (79%) and lowest among males with CLP (37%). Similar findings emerged on other indicators of academic progress, including grade retention and below-grade performance on individually administered achievement tests. These findings are in stark contrast to population-based epidemiological studies, which suggest that roughly 10–20% of the population have a learning disability (e.g., Altarac & Saroja, 2007; Rutter, 2004). Unfortunately, Broder et al. (1998) did not present data to characterize the nature of the learning problems observed (e.g., specific areas of academic weakness). In a recent population-based study, Yazdy, Austry, Honein, & Frias (2008) found that children with nonsyndromic clefts were roughly three times as likely to have received special education services as unaffected controls. This difference was most apparent for those with CLP (prevalence ratio = 3.3) and lowest for those with cleft lip only (CL; prevalence ratio = 1.7). Speech–language therapy was the most common service received, though rates of referral were also higher for other service categories (e.g., learning disabled and severe behavioral disorders).

Richman and colleagues have published several studies suggesting that reading delays are particularly common among children with orofacial clefts, at least in relation to test norms. For example, Richman, Eliason, and Lindgren (1988) found that children with clefts scored below-grade level on measures of word recognition and reading comprehension. Subgroup analyses revealed that this effect was most apparent at younger ages (i.e., ages 6–7 years) and among children with CP. Among older children (ages 10–13 years), word recognition scores were roughly grade-appropriate. However, those with CP continued to score below expectation in reading comprehension. The authors hypothesized that at younger ages, peripheral language problems (e.g., difficulty with phonological processing) might have accounted for the higher rate of reading problems among children with CLP and CP. Among older children with CP, continuing problems with reading comprehension were thought to reflect central language dysfunction. Other studies by Richman (e.g., Richman and Eliason, 1984) have found that children with clefts scored lower than expectations based on test norms.

More recent studies (e.g., Richman and Ryan, 2003; Richman, Wilgenbusch, & Hall et al., 2005) have examined the nature of these reading problems. Among children with clefts, those with reading disabilities are differentiated from nondisabled readers on the basis of verbal fluency and rapid naming but not phonemic awareness (Richman & Ryan, 2003). Richman et al. (2005) found that children with clefts had particular difficulty with a short-term memory task requiring them to point to a visually presented target after a brief delay. Children with deficits on this task were more likely than those without deficits to exhibit reading disabilities (61% vs. 30%). The investigators concluded that children with clefts might fail to spontaneously apply verbal labels for visual stimuli to aid memory. Therefore, they proposed that reading disabilities in this population are characterized by a form of “dysnomia”, or inefficient verbal labeling of visual stimuli, that might require different interventions than the phonological processing interventions used with dyslexic noncleft children.

Although these findings point to potentially important differences in learning among children with isolated orofacial clefts, virtually all of the published reading studies have compared children with clefts to test norms rather than recruiting samples of demographically similar controls. This is an important limitation, as samples recruited through hospital-based programs may differ in multiple ways from those included in test norms (e.g., over- or underrepresentation of families from low socioeconomic backgrounds), making the validity of the norms questionable. In one of the first studies to include a control sample (Collett, Leroux, & Speltz, in press), we examined language and reading outcomes in children with CLP and CP relative to a demographically similar control group. Participants in this study were recruited in infancy (age 3 months) and followed up to age 7 years. Outcome measures included assessments of expressive and receptive vocabulary, as well as measures of basic reading from the Woodcock–Johnson Achievement Battery - Revised (WJ-R; Woodcock & Johnson, 1989). Differences in expressive and receptive language were small and not statistically significant. On reading tasks, children in both the case and controls groups scored within the average range compared to test norms. When examining group differences, children with CLP did not differ from controls and those with CP actually scored higher than controls on academic achievement measures. A limitation of this study was that we did not measure other functions known to be associated with reading, such as phonological processing, phonological memory, and rapid automatized naming. These skills may be more sensitive to group differences than measures of basic reading, particularly in a sample
of relatively young readers. Further, as this is the first published study on the topic that failed to find reading deficits and the sample was relatively small (n = 41 cases, n = 53 controls), the findings warrant replication.

In the present study, we sought to test the hypothesis that children with orofacial clefts would score lower than demographically similar unaffected controls on measures of reading and closely related skills. This study builds on previous work by including a sample of unaffected, demographically similar controls for comparison, and by assessing language functions known to be highly correlated with reading among noncleft children (National Institute of Child Health and Human Development, 2000). Children in kindergarten through second grade were targeted for recruitment both because this has been found to be the time period of highest risk in prior studies by Richman and colleagues, and because this represents a critical period for identifying and intervening with reading problems (National Institute of Child Health and Human Development, 2000). Measures were chosen based on the dyslexia literature as well as the literature on reading problems among noncleft children with language delays (Raitano, Pennington, Tunick, Boada, & Shriberg, 2004; Velluntino, Fletcher, Snowling, & Scanlon, 2004) and other clinical populations (e.g., children with attention deficit hyperactivity disorder; Willcutt, Pennington, Olson, Chhabildas, & Hilslander, 2005). Specifically, we sought to assess known correlates of reading acquisition including phonological awareness, phonological memory, and rapid automatized naming. Our battery was designed to assess basic reading skills (i.e., single word and nonword reading), reading comprehension, and reading fluency.

Methods

Participants

Children with Orofacial Clefts

Children with nonsyndromic clefts of the lip only (CL, n = 8), lip and palate (CLP, n = 22), and palate only (CP, n = 12) were recruited from the Craniofacial Center at Seattle Children’s Hospital. Children with clefts were considered eligible if they were between the ages of 5 and 7 years at the time of approach, enrolled in kindergarten through second grade, and English was the primary language spoken at home. Exclusions included the presence of a known genetic or neurodevelopmental syndrome (e.g., 22q11 deletion syndrome, Stickler Syndrome, fetal alcohol spectrum disorder), visual and auditory impairments that would preclude participation (e.g., deafness/blindness), previous diagnosis of mental retardation, history of traumatic brain injury, and out-of-home placement (e.g., foster care). Initially, all parents of potentially eligible children were identified from the center’s patient registry and approached in writing with information about the study. Parents were asked to complete and return a response card indicating their willingness to be contacted about the study. Letters describing the study were initially sent to the families of 154 children. Twenty-five of these families could not be located and four declined to be contacted further. Follow-up phone calls were made to those who expressed an interest in participating and nonresponders who resided within roughly 2 hr of the research center. Phone calls were made to 72 families, and 58 were successfully reached and screened. Fourteen families declined participation, and two children were determined ineligible (one due to mental retardation, one due to inadequate English proficiency). Forty-two children were ultimately determined eligible and consented for participation.

Controls

A sample of 43 unaffected controls was recruited through advertisements posted on local internet sites (e.g., Craigslist) and through a registry of potential research participants maintained by the University of Washington (without affiliation to Seattle Children’s Hospital). This registry includes families who, at the time of their child’s birth, filled out and returned a response card indicating a willingness to be contacted with information about research studies. Families are re-contacted when their child reaches the age of 3 years to update their information and to determine whether they continue to be willing to be contacted. Families from this registry were contacted by phone and provided with information about the study. All control families who expressed an interest in participating were screened by phone to confirm eligibility. In addition to the exclusions listed for children with clefts, controls were excluded if they had a history of any craniofacial anomaly (e.g., orofacial clefting and craniofacial microsomia). An effort was made to ensure that control participants were demographically similar to children in the cleft group in terms of gender, age, grade level, and socioeconomic status (SES; Hollingshead, 1975). Eight families responded to advertisements, and all were determined eligible and enrolled in the study. A total of 66 families from the participant registry were contacted by phone and approached for participation. Of these, six families declined to participate, three children were determined ineligible, and 22 were not enrolled because they were not a demographic match (all due to high SES). The remaining 35 children were determined eligible, and their parents consented to their participation.
Measures

Medical and Developmental History Interview
A brief, semi-structured interview was completed with the parents of participating children. Educational history questions addressed the child’s educational placement, history of special education and related interventions, and home literacy practices (e.g., shared oral reading). Because chronic otitis media and hearing impairment are common among children with clefts and may be related to language and reading, parents also reported on their child’s history of suspected and confirmed hearing problems, otitis media, and pressure equalization (PE) tube placement.

Basic Reading Skills
Selected subscales from the Woodcock-Johnson Achievement Battery - III (WJ-III; Woodcock, McGrew, & Mather, 2001) were used to assess basic reading. Letter-Word Identification is a measure of letter and single word reading that requires the child to identify a series of increasingly difficult words. Passage comprehension involves reading a series of brief passages and filling in a missing word. Finally, Word Attack requires the child to answer questions about letter sounds and to read a series of increasingly difficult nonsense words using phonemic cues. The normative sample for the WJ-III included a heterogeneous sample of 4,740 children/adolescents in Grades k-12. The psychometrics for the WJ-III are well supported. For children in the target age range for this study, test–retest reliabilities range from \( r = .79 \) to \( .92 \). In addition to being widely used in the research literature, the WJ-III is one of the standard measures used in school systems across the country to determine children’s eligibility for special education, lending support to the external validity of our findings. Standard scores (using age-based norms) were used for analyses.

Reading Fluency
The Test of Word Reading Efficiency (TOWRE; Torgesen, Wagner, & Rashotte, 1999) was used as a measure of reading fluency for participants aged 6 years and above (i.e., those for whom normative data are available). The TOWRE involves reading a series of increasingly difficult sight words or nonsense words aloud. Scores are based on the number of words read correctly in 43 s. The TOWRE was normed with a sample of 1,507 individuals aged 6–25 years. For children aged 6–7 years, internal consistencies for the TOWRE range from \( .90 \) to \( .98 \), and test–retest reliabilities range from \( .90 \) to \( .97 \). The validity of the TOWRE is supported by convergence with similar measures and discrimination of clinical groups. Age-based standard scores for the TOWRE total composite were used for analyses.

Phonological Awareness and Phonological Memory
The Phonological Awareness and Phonological Memory scales from the Comprehensive Test of Phonological Processing (CTOPP; Wagner, Torgesen, & Rashotte, 1999) were used to measure these skills. The Phonological Awareness scale includes subtests requiring respondents to break down a series of words and nonwords into component sounds (Elision), to blend sounds to form whole words (Blending Words), and to identify beginning and ending sounds (Sound Matching). Phonological Memory includes nonword repetition and digit recall tasks. The CTOPP was normed with a diverse sample of 1,656 participants aged 5–21 years. The psychometrics of the CTOPP are excellent (e.g., internal consistencies = .86–.96, test–retest reliability = .79–.88), and validity is well-established.

Rapid Serial Naming
The Letter and Digit Naming subtests from the RAN/RAS (Wolf & Denckla, 2005) were used to assess rapid serial naming. These tasks involve a child naming aloud, as quickly as possible, a series of numbers or letters. Scores are based on the time elapsed. The Rapid Automated Naming/Rapid Alternating Stimulus Test (RAN/RAS) was normed with a sample of 1,461 children and adults. Test–retest stability for the RAN/RAS is good to excellent for school-age children (\( r = .81–.91 \)). Deficits in rapid naming are thought to reflect difficulty with the efficient retrieval of information from memory and production of a verbal response. Rapid naming of alphanumeric symbols (i.e., letters and numbers) has been found to be highly correlated with reading skill (Wolf, Bowers, & Biddle, 2000). For this study, a composite was generated by averaging children’s standard scores on the letter and number naming subtests.

Procedures
Child assessments were completed by trained psychometrists in accordance with standardized procedures, with breaks taken as needed. When scoring the various measures, children were not penalized for articulation errors. Auditory stimuli (e.g., for the CTOPP) were presented using a tape recorder with volume set according to child preference. Child testing sessions were completed in 1.5–2 hr. Child assessments were completed in clinic rooms with a one-way mirror, and videotaped for later review if needed for scoring. Participants received monetary reimbursement for their time ($25), and children...
received a small toy. Parents were able to receive a letter summarizing their child’s test results if desired. Before participating in the study, written informed consent was obtained from all parents and verbal assent was obtained from participating children. All procedures were approved by the Institutional Review Board and were in full HIPAA compliance.

**Analyses**

Power analyses were calculated prior to the study to determine sample size. These analyses were run using an $\alpha$ of .02 to control for multiple comparisons and an effect size of $\geq 0.8$ for univariate comparisons. Additional precision, gained through demographic matching and adjustment of potential confounders, and the use of a “false discovery rate” approach (described subsequently) were expected to increase power to detect differences. Based on these analyses, we determined that a sample of 36 cases and 36 controls would result in adequate power (i.e., $\geq 80\%$). We were ultimately able to increase our sample size in both case and control groups, resulting in power of 91.3% to detect an effect of $\geq 0.8$.

To evaluate potential response bias among cases, we compared the demographics of our participants with those who appeared to be eligible but declined participation or failed to respond to recruitment efforts (nonparticipants). Frequencies and $\chi^2$-analyses were used to compare participants and nonparticipants in terms of gender distribution, ethnicity (Caucasian vs. non-Caucasian), and Medicaid status (used as a proxy for SES). Demographic and clinical characteristics were summarized for cases and controls using descriptive statistics and frequencies, as well as $\chi^2$-analyses (for categorical variables) or t-tests (for continuous measures). To evaluate whether children with orofacial clefts scored lower than demographically similar unaffected controls on measures of reading and related skills, we performed a series of linear regression analyses comparing children with clefts to controls on scores for age-standardized basic reading skills, reading fluency, phonological awareness and memory, and rapid serial naming. Although the case and control groups were well-matched, we controlled for demographic and environmental variables known to be associated with reading skill to increase precision in our analyses. These potential confounds were identified *a priori*, and included age at assessment (continuous), sex, months of school (continuous), SES (measured using Hollingshead’s continuous score), and minutes per week of shared oral reading (continuous). In secondary analyses, we added history of suspected or confirmed hearing problems to determine whether hearing might account for observed differences. We also considered adjusting for PE tube placement, a possible marker of hearing loss due to repeated otitis media. However, given the desire to aggressively manage otitis media among children with clefts, PE tubes are placed in the majority of cases, sometimes proactively, and were considered an imprecise measure of hearing impairment. We also examined descriptive differences for cases with and without a history of special education or other interventions targeting reading. Due to small sample sizes by cleft type, we were not able to examine differences among children with CL, CLP, and CP.

We interpreted case–control differences in relation to the “false discovery rate” (FDR; Benjamini & Hochberg, 1995), which adjusts for the increased probability of Type I errors associated with multiple comparisons. Observed $p$-values are ordered from smallest to largest, and a $p$-critical value is derived for each comparison. $P$-values below the $p$-critical are considered statistically significant. As a preliminary study of reading and related skills, we considered FDR to be appropriate and this allowed us to examine multiple aspects of reading to avoid “missing” a potentially important group difference.

All statistical analyses were conducted using Stata statistical software (version 10.0, Stata Corp., College Station, TX).

**Results**

Data to evaluate response bias were incomplete in some cases for nonresponders (e.g., ethnicity data were missing for 17.5%, Medicaid data were missing for 13.2%). Although not statistically significant, participants were more likely than nonparticipants to be male [66.7% vs. 59.7%, $\chi^2 (1, N = 156) = 0.64, p = .42$] and Caucasian [69.1% vs. 50.9%, $\chi^2 (1, N = 136) = 0.70, p = .41$]. More notably, nonparticipants were nearly three times as likely as participants to receive Medicaid coverage [35.4% vs. 11.9% among participants; $\chi^2 (1, N = 141) = 7.98, p = .005$], suggesting that lower SES families were underrepresented in our sample.

Demographic characteristics for the two samples are summarized in Table I. Children with and without clefts were similar in terms of age [$t (83) = 0.16, p = .88$], sex [$\chi^2 (1, N = 85) = 0.03, p = .88$], months of school [$t (83) = -0.70, p = .49$], and minutes of shared oral reading per week [$t (82) = -1.03, p = .31$]. SES was slightly higher among families in the control sample [$t (82) = 1.13, p = .26$], as was the proportion of Caucasian participants [$\chi^2 (1, N = 85) = 2.36, p = .12$].
Otis media was common among both children with clefts and controls \( \chi^2(1, N = 85) = 3.44, p = .06 \); however, children with clefts were much more likely to have received PE tubes \( \chi^2(1, N = 85) = 41.78, p < .001 \). Hearing problems were more common among children with clefts \( \chi^2(1, N = 85) = 10.47, p = .001 \). Children with clefts were more likely to have been evaluated for special education \( \chi^2(1, N = 85) = 16.28, p < .001 \). Though not statistically significant, cases were also more likely to have received additional educational support in reading \( \chi^2(1, N = 85) = 3.25, p = .07 \).

As seen in Table II, mean scores for children with clefts and controls were within the average range on all measures relative to test norms. After adjusting for potential confounds, children with clefts scored significantly lower than controls in single word and nonword reading \( p = .007 \) and \( .003 \) on the WJ-III Letter–Word and Word Attack subtests, respectively), reading comprehension \( p = .001 \) on the WJ-III Passage Comprehension subtest), reading fluency \( p = .002 \) and .01 on the TOWRE Sight Word and Phonemic Decoding subtests, respectively), and nonword repetition \( p = .001 \) on the CTOPP NonWord Repetition subtest). Differences were robust in each of these areas, with standardized mean difference effect sizes ranging from \(-0.60 \) to \(-0.77 \) on the WJ-III scales and TOWRE, and an effect size of \(-0.73 \) on the CTOPP NonWord Repetition task. Differences on the RAN/RAS were not statistically significant \( p = .565 \). Similarly, although children with clefts scored lower than controls on all but one of the phonological awareness tasks from the CTOPP, these differences were not statistically significant with \( p \)-values ranging from .033 to .909, and all considered nonstatistically significant after FDR correction.

In analyses adjusting for a history of suspected or confirmed hearing problems, the magnitude of group differences was reduced slightly for most measures (Table III). Differences on the WJ-III Letter–Word Identification and the TOWRE Phonemic Decoding subtests were no longer statistically significant after adjusting for multiple comparisons \( p = .03 \) and \( .02 \), respectively). Differences on the WJ-III Word Attack and Passage Comprehension subtests \( p = .007 \) and \( .002 \), respectively), the CTOPP NonWord Repetition \( p = .004 \), and the TOWRE Sight Word \( p = .012 \) subtests all remained statistically significant.

Because children with clefts were more likely to have been referred for educational intervention, we were interested in determining whether these services might have reduced case–control differences. We therefore examined differences in reading scores between cases with no history of special education or reading intervention and those who had been evaluated for special education or received additional services in reading (results not shown). In all areas, children who had been evaluated for special education or received additional services in reading scored lower than other cases.

### Discussion

These findings are consistent with previous studies of reading and academic achievement among children with orofacial clefts (e.g., Richman et al., 1988; Yazdy et al., 2008) and provide support for the hypothesis that children with orofacial clefts score lower than controls on measures of basic reading and nonword repetition. These differences were statistically significant and moderate in magnitude, with mean difference effect sizes ranging from 0.60 to 0.74. Group differences were reduced in analyses adjusting for hearing problems, though still statistically significant.
for most outcome measures. Together, these findings suggest that the increased incidence of hearing and related problems may at least partially account for differences observed in reading and related skills. Further, children in the cleft group were more likely to have been evaluated for special education services and to have received additional support in reading, suggesting that many had already come to the attention of teachers and other school professionals. Although these children scored lower than cases who had not received such evaluation or intervention, these educational services may have attenuated case–control differences via improvements in language and early reading.

Phonological memory, assessed by the Nonword Repetition task from the CTOPP, appears to be an area of relative weakness for children with clefts. Phonological memory is thought to be important as children learn new and increasingly complex words that require them to sound out the component parts (Wagner et al., 1999). Additionally, there is some evidence that deficits in this area remain even after successful reading intervention (Bishop & Snowling, 2004). Group differences were not observed on other types of phonological tasks from the CTOPP, suggesting that they had achieved these basic skills or that these skills had been remediated with intervention. However, children with clefts did struggle with tasks requiring the application of these skills, such as the Word Attack subtest from the WJ-III and the phonemic decoding subtest from the TOWRE. Differences between children with and without clefts may become increasingly apparent as the demands of reading increase, moving from single word decoding to sounding out unfamiliar words and reading for meaning.

The findings of this study contrast with those from an earlier investigation (Collett et al., in press), in which we failed to find significant differences between children with clefts and controls using comparable reading subtests from the WJ-R. In both the earlier and current study, children with clefts scored within the average range compared to the WJ-R. Differences between children with and without clefts may at least partially account for differences observed in reading and related skills. Further, children with clefts did struggle with tasks requiring the application of these skills, such as the Word Attack subtest from the WJ-III and the phonemic decoding subtest from the TOWRE. Differences between children with and without clefts may become increasingly apparent as the demands of reading increase, moving from single word decoding to sounding out unfamiliar words and reading for meaning.

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had a greater interest in research or intellectual curiosity, characteristics likely to be evident in other ways in the home (e.g., greater emphasis on education). These findings highlight the importance of future studies with larger samples, with careful attention given to potential sources of ascertainment bias among both children with clefts and controls.

Our findings, in combination with the existing literature, suggest that larger scale studies are warranted to clarify the etiology of reading differences among children with clefts. Recent work by Nopoulous (e.g., Nopoulous et al., 2000, 2002) and others (e.g., Ceponiene et al., 2000) suggests the possibility of neurobiological differences among individuals with clefts, particularly in the temporal lobe and other regions associated with language and reading skills. The orofacial cleft may therefore be a “marker” of aberrant embryonic development, affecting both the face and the brain. However, the alternative hypothesis that differences are explained by the sequelae of clefting has not been well tested. Further, these hypotheses need not be mutually exclusive—for example, children with clefts may have initial neurobiological vulnerabilities that are exacerbated by their hearing and speech impairments. Ideally, future studies would involve reading measures comparable to those administered here along with direct measures of neurobiological function (e.g., fMRI or ERP), speech, and hearing at multiple points in time, providing developmental data to help clarify etiological hypotheses.

A few other limitations and directions for future research warrant discussion. Our analyses of demographic characteristics for participants versus nonparticipants suggest that children from lower SES backgrounds were underrepresented in this study. This is an important limitation, as reading and language are known to correlate with SES (Noble, McCandliss, & Farah, 2007). This would not affect our comparisons with unaffected controls, who were demographically similar to cases, but children from lower SES backgrounds might be expected to score lower compared to test norms. In fact, the combination of low SES and a medical condition such as an orofacial cleft may be particularly detrimental, as such families may have reduced access to medical care and other needed services (e.g., speech and language therapy) and may be less able to advocate for their child’s education. A second limitation relates to our assessment battery, which did not include direct measures of articulation or hearing. Our data suggest that parent-reported hearing problems were associated with reading outcomes, though direct assessment of hearing would have provided a more reliable and sensitive assessment of this effect. Finally, our sample was insufficient for subgroup comparisons (e.g., by grade level, sex, cleft type, etc.). Differences by cleft type are of particular interest, as this may help to clarify the etiology of reading problems in this population. For example, because CL and CLP are thought to share a common etiology, a direct effect of clefting on brain development would be expected to result in similar scores on neuropsychological measures. If reading problems are attributable primarily to early speech or hearing impairment, one would expect similarities between children with CLP and CP and the highest scores should be observed in children with CL, who typically have little or no speech impairment and fewer episodes of otitis media.

Clinically, these findings support routine neuropsychological screening of children with clefts by cleft/craniofacial teams, as advocated by the American Cleft Palate Craniofacial Association (ACPA, 2003). Through their frequent contact with children with clefts and their families, cleft/craniofacial teams have the opportunity to identify children with heightened risk for reading delays before they fail in school and fall behind their peers.
Optimally, this would result in earlier identification and remediation of reading problems to avoid adverse outcomes. In our sample, many children had already come to the attention of school personnel. This may be due to close monitoring by the craniofacial team and the relatively high SES of participating families, who were likely attuned to their child’s academic performance. In lower SES populations and among other groups that may be less likely to advocate for school services, advocacy by providers on the cleft/craniofacial team may be particularly important in raising awareness of early learning and academic resources. Richman et al. (2005) have suggested that children with clefts might require different interventions than those validated with noncleft children who struggle in reading, and our failure to find differences on several of the language tasks thought to be precursors of reading (e.g., phonemic decoding, rapid naming, etc.) may lend some support to this hypothesis. That is, while they were performing below their peers in single word reading and reading comprehension, children with clefts did not receive lower scores on tasks measuring these component skills. Ultimately, it may be that the predictors of reading acquisition differ in this group and that targets for intervention need to be adjusted accordingly. This notion requires further study, potentially using a “response to intervention” approach to document the rate of reading acquisition for children with and without orofacial clefts when provided with well-validated teaching methods. At present, routine clinical assessment and monitoring of reading in this population appears warranted. Until further data become available on potentially unique predictors of reading in this population, preschool-age screening might include assessments of letter knowledge, phonemic awareness, and phonological memory. Because there is a risk of “missing” children at risk using these assessments, ongoing monitoring of reading acquisition and the development of more advanced reading skills (e.g., reading efficiency, comprehension, etc.) during the early elementary school years are also recommended.

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