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<LRH>*Jessica O. Boyce* et al.

<RRH>*Language in children with cleft lip and/or palate*

Research Report

Receptive and expressive language characteristics of school-aged children with non-syndromic cleft lip and/or palate

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Abstract

Background: Research investigating language skills in school-aged children with non-syndromic cleft lip and/or palate is sparse. Past studies focus on younger populations, lack key comparisons to demographically matched control cohorts or explore language as a component of broader academic skills. Trends of existing studies suggest that affected children may perform at a lower level compared with typically developing peers.

Aims: To examine the receptive and expressive language skills of middle-school-aged children with non-syndromic cleft lip and palate (CLP) and cleft palate only (CP). Additionally, to explore the language skills of children with clefts compared with a non-cleft control group.

Methods & Procedures: Thirty-seven participants with orofacial clefts (aged 7;1–14;1 years) participated in the study: 19 with CLP (10 males; 9 females) and 18 with CP (8 males; 10 females). A non-cleft comparison group consisted of 129 individuals matched on age, sex and maternal education level. Participants completed formal language (Clinical Evaluation of Language Fundamentals, Fourth Edition) and non-verbal intellectual measurements (Wechsler Abbreviated Scale of Intelligence—WASI). Demographic and developmental information was obtained via parental interview. Further clinical details (e.g., surgery; hearing status) were extracted from patient medical files. Cleft and non-cleft language and non-verbal IQ outcomes were reported separately. Language outcomes were then compared between groups.

Outcomes & Results: Participants with clefts achieved core (mean = 103.31, standard deviation (SD) = 10.31), receptive (mean = 102.51, SD = 11.60) and expressive (mean = 102.89, SD = 12.17) language index scores within the normative average range. A total of 14.1% and 17.8% of the cleft and non-cleft groups respectively had impairment (i.e., ≥ 1.25 SD below the mean) in one or more language domains. No significant differences were found in the three language index scores between cleft and non-cleft groups.

Conclusions & Implications: This study is the first formally to examine language skills alongside non-verbal IQ in school-aged children with clefts compared with a large matched non-cleft population. Results suggest that health professionals should evaluate each child as they present and not assume that a child with non-syndromic CLP or CP will also have co-occurring language difficulties. Where language falls in the average range, these skills can be harnessed to support areas of difficulty often associated with orofacial clefting, such as speech.

Keywords: non-syndromic, cleft palate, cleft lip and palate, expressive language, receptive language.

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<A>**What this paper adds**

<B>*What is already known on the subject*

Children with non-syndromic cleft lip and/or palate (CL/P) often show delays in early babble, lexical and syntactic development. Studies suggest that difficulties persist through school years when children with clefts have poorer academic and cognitive development than their peers. Yet there is a lack of knowledge surrounding the receptive and expressive language abilities of children with non-syndromic CL/P during the middle-school years.

<B>*What this paper adds to existing knowledge*

This study provides a standardized evaluation of language skills amongst school-aged children with non-syndromic CLP and CP compared with a demographically similar non-cleft group. Children with clefts had mean receptive and expressive language abilities within the normative average range and similar to their non-cleft peers. Language impairment was present in both groups at similar proportions: 14.1% of the cleft group and 17.8% of the non-cleft group.

<B>*What are the potential or actual clinical implications of this work?*

Health professionals should evaluate each child as they present and not assume that a child with non-syndromic CLP or CP will also have co-occurring language difficulties. Where language falls in the average range, these skills can be harnessed to support speech difficulties often associated with orofacial clefting.

<A>**Introduction**

The term 'non-syndromic cleft lip and/or palate' (CL/P) comprises a group of orofacial anomalies resulting from disrupted gestational tissue growth. Following surgical repair, persisting structural, hearing and neurocognitive deficits may lead to communication difficulties that impact learning, relationships and psychosocial well-being (Wehby *et al.* 2014, Richman *et al.* 2012). While much is known about the speech features of individuals with CL/P, associated language development is not widely explored.

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<B>*Language development in toddler and pre-school children with CL/P*

To date, investigations of language skills have focused on toddler, infant and pre-school groups, primarily through parent report and informal language sampling (Hardin-Jones and Chapman 2014, Broen *et al.* 1998). Children born with cleft lip and palate (CLP) or cleft palate only (CP) may show limited complexity in their speech and expressive language from first vocalizations (Hardin-Jones and Chapman 2014). Toddlers up to 24 months of age with non-syndromic CLP or CP also use less lexical items when compared with typically developing peers, likely due to a small accompanying consonant inventory (Chapman *et al.* 2003, Broen *et al.* 1998). Those with CL/P may continue to perform poorly on standardized expressive language assessments and measures of utterance length up to 30 months of age (Scherer and D'Antonio 1995, Jocelyn *et al.* 1996). On the other hand, receptive language outcomes amongst similar age groups are generally within typical limits (Scherer and D'Antonio 1995, Snyder and Scherer 2004). The few studies reporting early receptive delays suggest hearing loss as a key associated factor (Broen *et al.* 1998, Jocelyn *et al.* 1996).

<B>*Language development in primary school-aged children with CL/P*

A handful of studies have explored receptive and expressive language skills in primary school-aged groups, and results are inconclusive. Children with CLP or CP aged 3–10 years have been shown to demonstrate language abilities that are comparable with typically developing age-matched peers (Collett *et al.* 2010a, Chapman *et al.* 1998, Chapman 2011) and standardized test norms (Feragen *et al.* 2017, Morgan *et al.* 2017). However, Morgan *et al.* (2017) noted that a high proportion (20%) of children had impairment in at least one area of language (i.e., overall, receptive or expressive), with different patterns for receptive (9.1%) and expressive delays (18.2%). Contrasting results were reported by a further study, however, where a group of 16 Persian-speaking children with CLP performed significantly worse than a normative population on the Persian Test of Language Development—Primary, 3rd edn (Anaraki *et al.* 2016). Small sample sizes and the lack of non-cleft comparison groups in a number of these studies disregard the impact of crucial demographic factors (e.g., socioeconomic background), making their observations difficult to generalize.

<B>*Population-based studies of language in children with CL/P*

Large-scale studies of over 500 participants with clefts observe that those with CL/P have weaker academic skills when compared with non-cleft peers (Wehby *et al.* 2014, Bell *et al.* 2017). In an American population-based study, children from primary to high school with CP scored significantly

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below their classmates across all domains of academic achievement—reading, language and mathematics—while those with CLP performed significantly poorer in all areas apart from reading (Wehby *et al.* 2014). A recent Australian study found that children with CP aged 8–14 years of age were less likely to meet minimum national standards for reading, grammar and punctuation than their peers (Bell *et al.* 2017). However, when all cleft types were considered together, over 80% reached the national minimum standards. Further, a meta-analysis by Roberts *et al.* (2012) also described overall cognitive functioning as an area of difficulty amongst children and adults with CL/P, with most significant deficits found in language. While these large-scale studies provide valuable population-based data, they examine language as part of broader assessments and lack linguistic complexity and detail. That is, there is no examination of performance on specific sub-domains of language, i.e., language expression versus comprehension, or on domains of morphology, syntax or semantics, areas that have previously been shown to differ amongst this population (Morgan *et al.* 2017, Scherer and D'Antonio 1995).

<B>*Factors associated with language development*

Specific language impairment, now more commonly reported as developmental language disorder (Bishop 2017), is often associated with impaired non-verbal IQ; this being documented in both CL/P samples (Roberts *et al.* 2012, Richman 1980) and broader population groups (Botting 2005, Conti-Ramsden *et al.* 2012, Gallinat and Spaulding 2014). Conti-Ramsden *et al.* (2012) observed that non-verbal skills co-varied with language ability in a large group of 11-year-old children with history of specific language impairment. A later meta-analysis of 131 studies expanded upon these findings and reported that non-verbal IQ was an average of [AQ2] 0.75 SD (standard deviation) lower for children with language impairment when compared with typically developing peers (Gallinat and Spaulding 2014). Unfortunately, within the relevant cleft literature, language data are generally extracted from IQ assessments and therefore do not provide a representative estimate of receptive and expressive language skills. Moreover, studies that do differentiate language domains often fail to address non-verbal IQ.

Cleft type is well documented to affect speech outcomes, with an arguably more severe cleft (i.e., CLP) being associated with increased hypernasality and cleft related articulation characteristics (Schönweiler *et al.* 1999). Early speech difficulties have, in turn, been moderately correlated to subsequent vocabulary development (Chapman *et al.* 2003). Differences across cleft type continue into the primary school years, where those with CP are more likely to have global language deficits or learning disabilities while expressive difficulties alone are more commonly found in children with

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CLP (Broder *et al.* 1998, Richman 1980). These observations are in line with broader genetic and epidemiological studies where cleft lip with or without cleft palate and CP are often considered as distinct entities based on their different embryological origins (Fogh-Andersen 1942), recurrence risks (Sivertsen *et al.* 2008), genetic aetiologies and associations with underlying syndromes (Dixon *et al.* 2011). Nonetheless, in older and larger population-based groups, overall academic performance (including language) generally does not differ significantly with cleft type, although when compared with classmates, differences are generally largest for those with CP (Wehby *et al.* 2014, Feragen *et al.* 2017, Bell *et al.* 2017).

Speech and language skills may also be influenced by additional mediating factors including: a syndrome or developmental delay (Feragen *et al.* 2017); hearing impairment (Hall *et al.* 2017); sex (Persson *et al.* 2018); and maternal education and birth order (McKean *et al.* 2017). A higher level of maternal education along with being first born are linked with better performance on standardized language assessment measures amongst non-cleft populations (McKean *et al.* 2017). Furthermore, otitis media with effusion and associated conductive hearing loss are common amongst individuals with CL/P and have been linked to poorer receptive and expressive language skills (Schönweiler *et al.* 1999, Hall *et al.* 2017).

There remains a lack of understanding around the precise receptive and expressive language abilities of school-aged children with CL/P and their interaction with non-verbal intelligence. This is the first study formally to evaluate language skills alongside non-verbal IQ in children with non-syndromic CLP and CP during the middle-school years. It adds to existing work in the area by including a large non-cleft comparison group. The first aim was to examine the receptive and expressive language skills of middle-school-aged children with non-syndromic CLP and CP using a standardized assessment tool. The second aim was to explore the language skills of children with clefts (CLP and CP) compared with a demographically similar non-cleft control group. Exploratory analyses also examined any differences in language outcomes between the two cleft groups (CLP and CP).

<A>**Methods**

<B>*Participants*

Participants with non-syndromic CLP and CP were recruited between 2009 and 2010 from the Cleft Registry Database at the Royal Children's Hospital in Parkville, Victoria, Australia. Participants were included if they met the following criteria: (1) no syndromic diagnosis, abnormal DNA findings or

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obvious clinical syndromic features; (2) no co-occurring major medical conditions likely to impact speech or language development (e.g., epilepsy or traumatic brain injury); (3) normal hearing in at least one ear and no more than 50 dB loss in the other; and (4) English spoken as a first language. Participants' medical histories and genetic files were reviewed by a medical geneticist specializing in orofacial clefting to confirm a non-syndromic diagnosis. Demographic and developmental data were obtained through parental interviews and clinical information (e.g., surgery, hearing status) were extracted from patient medical files. A total of 118 participants were contactable and eligible for inclusion. Of these, 42 were recruited and agreed to participate and 37 (aged 7;1–14;1 years) completed the entire test battery.

The study was approved by the Human Research Ethics Committee of the participating hospital. Parents also provided written consent for their child's study participation.

A non-cleft comparison group was randomly selected from the Early Language in Victoria Study (ELVS) cohort. The ELVS is a population-based longitudinal study tracking language development in Melbourne, Australia (Reilly *et al.* 2018). In line with existing literature in this area, non-cleft participants were included if they met the inclusion criteria highlighted for those with clefting and did not have any history of craniofacial anomalies (Collett *et al.* 2010b, Chapman *et al.* 1998). Non-cleft participants were matched on age, sex and maternal education, factors considered to be important in language development (McKean *et al.* 2017, Norbury *et al.* 2017).

Demographic variables and cleft-related background characteristics are shown in table 1. While children with autism and attention-deficit hyperactivity disorder (ADHD) were not explicitly excluded, no included participants had autism and one participant had ADHD in both the cleft and non-cleft groups. The level of maternal education ranged from not completing school to obtaining postgraduate degrees, with most mothers completing high school. Of the children with clefts, nine received secondary surgery for velopharyngeal insufficiency. Seven participants had palatal fistula repairs and four with CLP had lip re-repairs. Speech data obtained within 6 months of the language assessment were available for 18 participants and showed that three had hypernasal speech and two had audible nasal air emissions. Nine participants had accompanying anterior oral cleft speech characteristics (e.g., lateralization or palatalization) and an additional three had posterior (e.g., backed to velar or uvular) or non-oral (e.g., glottal or pharyngeal articulation) errors with or without anterior errors. A total of 20 participants with clefts had received at least 6 weeks of speech therapy before assessment for this study.

<tab 1>

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<B>*Procedures and measures*

All participants completed standardized language and non-verbal IQ assessments in a quiet room in one sitting. Tests were administered and scored according to relevant test manuals.

Language was measured using the standardized Australian version of the Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4; Semel *et al.* 2003). CELF-4 is a widely used and globally validated clinical tool that enables comprehensive standardized assessment across all language domains (Eadie *et al.* 2014).

CELF-4 subtests were administered to generate composite index scores corresponding to each participant's age: Core Language Score (CLS), Receptive Language Index (RLI) and Expressive Language Index (ELI). The CLS reflects overall receptive and expressive language skills. Individual subtests assessed varying receptive and expressive domains including semantics, morphology and syntax. The following subtests were completed by all participants: Concepts & Following Directions, Recalling Sentences, Formulated Sentences and Word Classes—Receptive. Participants aged 8 years and below also completed the Word Structure and Sentence Structure subtests, while participants aged 9 years and above only completed the additional Word Classes—Expressive subtest. Subtests generated raw scores that were converted to norm-referenced scaled scores (mean = 10, SD = 3) and combined to generate the three composite scores (mean = 100, SD = 15) using the CELF-4 manual. Language impairment was defined as performance > 1.25 SD below the mean (Reilly *et al.* 2018). CELF-4 scores for the cleft group were calculated by the original test administrator and 100% were rated by a second individual (J.B.) to confirm scoring reliability. One participant with CLP had their core and receptive index scores subsequently excluded as two corresponding subtests had been administered incorrectly.

Non-verbal IQ was quantified using the WASI or WASI-II (2nd edn) and tested by psychologists or speech pathologists trained in administration of the tool (Wechsler 1999, 2011). Given the longitudinal nature of the ELVS, the updated WASI-II was used for a portion of the non-cleft participants. Non-verbal subtests Block Design and Matrix Reasoning were completed to generate a Performance IQ (PIQ) or Perceptual Reasoning Index (PRI) score for the WASI and WASI-II respectively (mean = 100, SD = 15). The PIQ and PRI have been shown to correlate well with each other—yielding a corrected correlation coefficient of 0.86 (Wechsler 2011)—and with full IQ batteries (i.e., Wechsler Intelligence Scale for Children—Fourth Edition; Wechsler 2003).

<B>*Statistical analysis*

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Descriptive statistics (frequencies, means, SDs, medians, interquartile ranges and proportions) were calculated and compared for demographic data and all standardized index and subtest scores. After matching on age, sex and maternal education, cleft and non-cleft comparisons for each language index were completed with multivariate linear regression. We also conducted exploratory analysis to examine any differences in language outcomes between the two cleft types using the following comparisons: CLP versus non-cleft; and CP versus non-cleft. A separate model was generated for each language index (CLS, RLI and ELI). In each model, we adjusted for differences in birth order and non-verbal IQ between groups; two variables important in language development that were not actively matched. To account for multiple comparisons, a Bonferroni significance threshold was applied and outcomes were considered to be significant when $p < 0.016$. Analysis was conducted using Stata 14.2 computer software.

<A>**Results**

The mean CLS from the CELF-4 for all participants with clefts was 101.58 (SD = 14.53, range = 41–112, n = 36), well within the standardized average range of 86–114 and just over the standardized mean of 100. Receptive language and expressive language indexes fell around the same position at 101.06 (SD = 14.40, range = 50–121, n = 36) and 101.43 (SD = 14.91, range = 49–122, n = 37) respectively. Mean non-verbal IQ (PIQ or PRI) was 102.86 (SD = 12.42, range = 75–129, n = 37), also within the standardized average range. The noticeably wide range in language index scores is due to an outlier in the group; a male with CP who achieved a score > 3 SD below the mean for all CELF-4 language indexes. He achieved a non-verbal IQ score of 75, [AQ3] 1.33 SD below the mean, indicating borderline non-verbal IQ. Figure 1 shows CLS graphed against non-verbal IQ, clearly identifying the outlier. This participant was excluded from remaining statistical analyses.

<fig 1>

<B>*Descriptive statistics*

With the outlier excluded, CELF-4 and WASI index and subtest group means for both cleft and non-cleft groups fell within the average range (table 2). The lowest scoring subtest for the cleft group was Sentence Structure (n = 8, standard score mean = 8.88, SD = 3.56), which examined the understanding of grammatical functions in spoken sentences of varying length and complexity.

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A total of 14.1% of individuals with clefts and 17.8% of the non-cleft group scored > 1.25 SD below the mean on one or more language index, indicating language impairment. Of these participants, receptive impairments were present for 8.6% (n = 3) of the cleft group versus 13.2% (n = 17) of the non-cleft group while 5.6% (n = 2) of the cleft group and 8.5% (n = 11) of the non-cleft group had impaired expressive language. Within these groups, only 3.9% (n = 5) of the non-cleft group had impairment in both receptive and expressive language. Figure 2 breaks down levels of impairment across receptive and expressive domains in both groups.

<fig 2, tab 2>

<B>*Comparison between cleft and non-cleft groups*

After adjusting for non-verbal IQ and birth order, no statistically significant differences between cleft and non-cleft groups were found across core, receptive and expressive language indexes (table 3). Similarly, no differences in index scores were found when stratifying models by cleft type (p > 0.05). As there were no differences in CELF-4 index scores, further analysis of subtests was deemed irrelevant.

<tab 3>

<A>**Discussion**

This study is the first to examine language skills in middle-school-aged children with orofacial clefts compared with a large matched non-cleft control group using a standardized clinical assessment tool. The primary aim was to examine the receptive and expressive language skills of children with non-syndromic CLP and CP. Observations were then compared with a demographically similar non-cleft group matched on age, sex and maternal education. Non-verbal intelligence (measured through the WASI and WASI-II) and birth order were also ascertained and adjusted for in analysis. Median and mean core, receptive and expressive language skills of children with clefts fell within average population norms and were comparable with the non-cleft group. The results add to the small pool of emerging evidence that formally examines language skills amongst younger pre- and primary school-aged children with CL/P (Collett *et al.* 2010a, Chapman 2011, Chapman *et al.* 1998).

Six participants with clefts had language impairment (≥ 1.25 SD below the mean) in one or more language domain. One of these participants was an outlier because he performed significantly lower than others on all three language indexes, widening the score ranges but not markedly

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impacting overall descriptive statistics. This individual was the only participant with a cleft to achieve a non-verbal IQ score > 1 SD below the mean, possibly contributing to his poor language scores. Despite not being diagnosed with a syndrome or intellectual disability, this participant had a history of ventricular septal defect, attended a special school and had a family history of learning disability. Taken as a whole, this history suggested a possible developmental delay not associated with any previously identified genetic diagnosis. For this reason, data from this participant were excluded from further statistical analyses.

<B>*Language compared with the standardized test norms*

Results from this study parallel existing literature, which suggests that children with non-syndromic CL/P generally perform within the average range on standardized language assessments (Collett *et al.* 2010a, Feragen *et al.* 2017, Morgan *et al.* 2017). Differences emerge when proportions of those with impairments are explored further. Morgan *et al.* (2017) reported that 20% of their participants who spoken English as a first language had an impairment (defined as a standard score of ≤ 80 on the CELF-4) in at least one language domain, greater than the 14.1% reported in this study. This overall difference is likely due to discrepancies in expressive (18.2% versus 5.6%) rather than receptive (9.1% versus 8.6%) impairment between the two studies. Similar comparisons with other studies are restricted by differences in assessment tools and methods of data exploration (Collett *et al.* 2010a, Feragen *et al.* 2017).

Variations found between this study and that of Morgan *et al.* (2017) may be explained by varied demographic factors between the two cohorts. While both studies examined children with non-syndromic CLP and CP, Morgan *et al.* studied a significantly younger cohort (mean age = 5.91 years), perhaps allowing for greater influences of speech difficulties and middle-ear dysfunction often more prevalent amongst younger age groups (Nyberg *et al.* 2014, Flynn *et al.* 2009). Other clinical and demographic factors (i.e., hearing status, speech therapy, birth order or maternal education) may have also contributed to outcomes; however, precise discrimination is challenging as different characteristics are reported across studies.

More meaningful observations can be made with a comparison with a demographically similar non-cleft cohort, as seen in this study. As expected, most participants in the non-cleft group achieved language scores within the average range. Proportions of those with language impairment in at least one domain were marginally lower for the cleft group (14.1%) when compared with the non-cleft group (17.8%). However, when examining results by level of impairment, where present, children with clefts consistently had greater proportions of difficulties (figure 2). While our inclusion

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of a non-cleft cohort allows for deeper exploration, similar studies with larger sample sizes are required to elucidate precise patterns of receptive and expressive language impairments.

Overall outcomes from this study differ from those investigating language as a component of broader academic or cognitive abilities (Roberts *et al.* 2012, Wehby *et al.* 2014, Bell *et al.* 2017). Such discrepancies may be explained by the nature of each assessment and its administrative context. The CELF-4 explores specific aspects of language in structured subtests, while broader academic and cognitive assessments examine a range of competencies through one task. The latter require a combination of skills including letter and sound knowledge, grammar, semantics, sequencing and memory. Deficits in any one of these areas may have contributed to the poorer results in previous studies. A benefit of the current study was the use of a language specific assessment tool that examined multiple language components, thus allowing for more detailed language characterization. Additional larger studies concurrently investigating standardized language and broader academic skills may help to elucidate accurately their relationship amongst this population.

<B>*Language compared with a demographically similar non-cleft group*

Results were similar when compared with a demographically matched non-cleft cohort. After adjusting for differences in potential confounding factors, we found small and insignificant differences across core, receptive and expressive language indexes. These results expand upon those previously reported by examining an older population and considering non-verbal IQ alongside a range of language domains including morphology, syntax, and semantic relationships (Chapman 2011, Collett *et al.* 2010a, Chapman *et al.* 1998).

Comparably, analysis showed no significant differences between those with CP versus controls and those with CLP versus controls. Whilst not a direct comparison, these findings contrast other American and Australian population-based studies of overall academic skills (including language, literacy and numeracy), where children with CP typically performed worse than those with CLP when compared with typically developing peers (Bell *et al.* 2017, Wehby *et al.* 2014). Nonetheless, in the current study, fewer than 20 participants were included in each cleft group. It is possible that greater differences may appear with a larger cohort.

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<B>*Strengths and limitations*

Methodologically, the addition of a large demographically similar non-cleft control group is a relative strength of this study. Existing work amongst similar populations generally examines language skills in relation to pre-published test norms, not allowing for interactions from potential mediating factors known to be important in language development (Anaraki *et al.* 2016, Feragen *et al.* 2017, Morgan *et al.* 2017). In this study, non-cleft participants were matched on age, sex and maternal education, while other potentially influencing factors—birth order and non-verbal IQ—were controlled for in the analysis. Additionally, our language assessment tool allowed for receptive and expressive language abilities to be standardized and reported separately.

Nonetheless, a number of limitations are worth noting. First, we did not have access to the speech profiles of all participants in the cleft group. Ideally, we would have considered measures of articulation alongside language to account for any interactions that may exist (Schönweiler *et al.* 1999, Chapman *et al.* 2003). Further, while participants with known hearing loss in both ears were excluded, comprehensive details of hearing status were not available for all included participants. However, given that participants with clefts performed similarly to those without, it is unlikely that hearing variations played a significant role within this cohort. Lastly, 31% of contactable participants completed the entire protocol for inclusion in this study. While comparison with a large non-cleft control cohort accounts for variations in demographic factors, potential selection bias does exist and results should be interpreted accordingly. The size of the CLP and CP groups was also insufficient for conclusions to be made based on cleft type, a comparison that may be considered in future studies and contribute to a greater understanding of this anomaly.

<B>*Clinical implications*

Clinically, the findings have positive implications for children with non-syndromic CLP and CP and their families. Where language falls within the average range, these skills can be harnessed to support broader areas of learning and development including speech, an area of difficulty often associated with orofacial clefting. The results suggest that, as a whole, language may not be an anticipated area of difficulty for children with non-syndromic CLP and CP. However, impairments may still be present and assessment of these skills should not be overlooked, particularly in the presence of an intellectual deficit. In clinical settings, health professionals should consider language abilities independently without making assumptions based on type of clefting or other clinical presentations. Finally, where an intellectual or language impairment is present later in life, it may be appropriate to revisit medical history and consider other relevant diagnoses.

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<A>**Conclusions**

Results from this study expand upon the limited body of existing evidence and suggest that middle-school-aged children with non-syndromic CLP and CP may have language skills within the average normative range compared with children without a cleft. Further, where impairments were present in one or more language domain, they were seen in similar proportions across cleft and non-cleft groups. Results add to the small number of language-focused studies in the cleft field by considering an older population of middle-school-aged children and through the inclusion of a demographically similar non-cleft cohort. Further research systematically exploring language sub-domains in larger populations is needed to substantiate these findings.

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Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

<<t/s Set names in caps and scaps as per usual style>>

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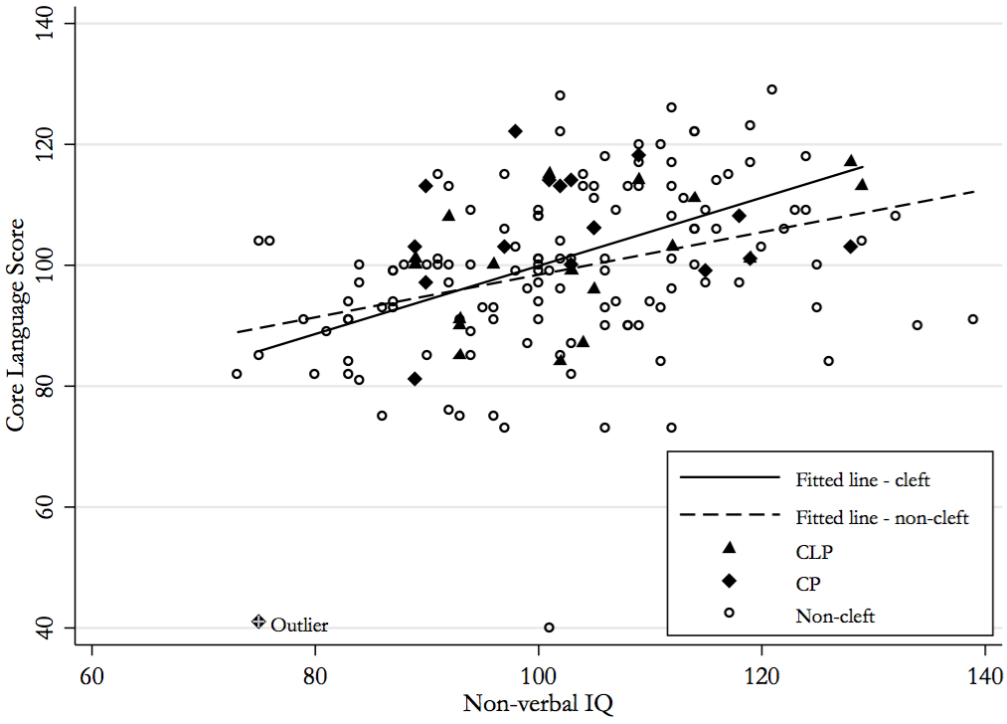
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Figure 1. Core language scores against non-verbal IQ. CLP, cleft lip and palate; CP, cleft palate only.



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Figure 2. Participants with language impairment (%).The identified outlier is excluded; percentages are calculated from different *n* values for receptive (*n* = 35) and expressive (*n* = 36) language indexes in the cleft group.

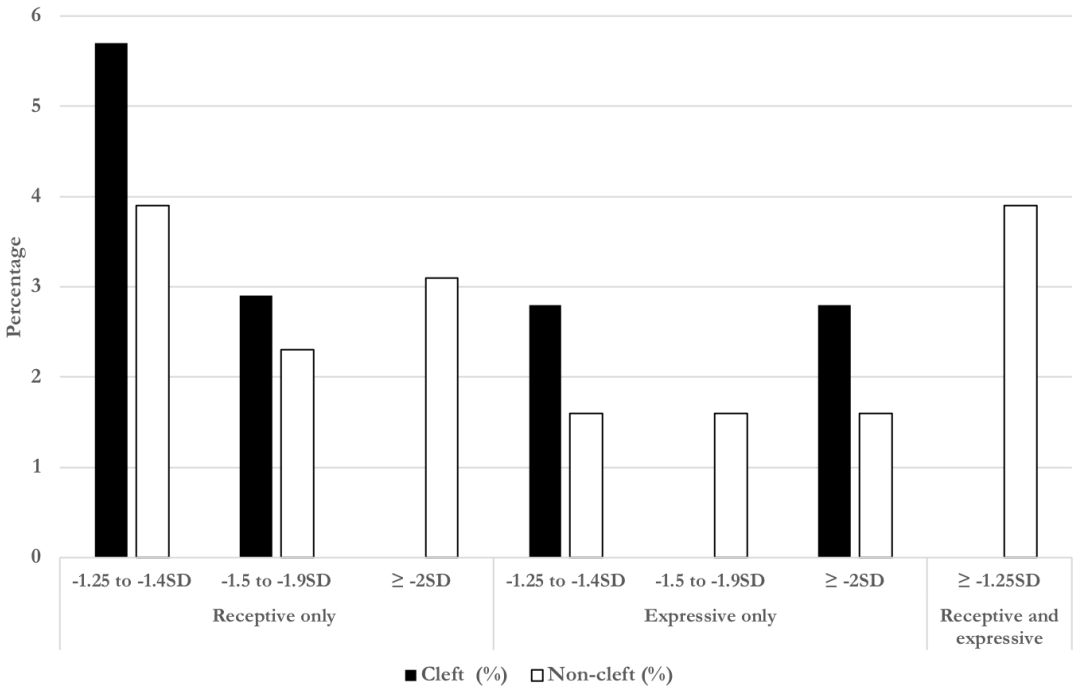


Table 1. Demographic and background information[a]

	CLP	CP	Non-cleft
Sex			
Female	9/19	10/18	70/129
Male	10/19	8/18	59/129
Age (years), mean (SD)	10.8 (1.9)	10.4 (1.9)	10.4 (1.8)
Birth order[b]			
First	3/19	11/18	69/129
Second	9/19	5/18	39/129

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Third	4/19	1/18	18/129
Fourth or later	3/19	1/18	3/129
Maternal education level			
< 12 years of schooling	5/18	3/18	17/129
12–13 years of schooling	11/18	5/18	64/129
Trade/apprenticeship	0/18	0/18	0/129
Certificate or diploma	0/18	5/18	20/129
Degree or postgraduate	2/18	5/18	28/129
Secondary procedure for VPI[c]			
Pharyngoplasty	3/19	3/18	0/129
Pharyngeal flap	1/19	0/18	0/129
Intravelar veloplasty	0/19	1/18	0/129
Fat graft	1/19	1/18	0/129
None	14/19	14/18	0/129

Notes: [a]Sample size is specified for each variable as some data are missing.

[b]Half siblings are included.

[c]The total for cleft palate only (CP) does not add up to 18 as one participant had both a pharyngoplasty and fat graft.

CLP, cleft lip and palate; SD, standard deviation; VPI, velopharyngeal insufficiency.

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Table 2. Description of CELF-4 and WASI index scores for cleft and non-cleft groups[a]

	CLP and CP			Non-cleft		
	n	Median (IQR)	Mean (SD)	*n*	Median (IQR)	Mean (SD)
CELF-4 indices						
CLS	35	103 (99–113)	103.31 (10.31)	129	100 (91–109)	99.13 (13.72)
RLI	35	103 (94–112)	102.51 (11.60)	129	97 (88–109)	97.50 (13.71)
ELI	36	104 (98–110)	102.89 (12.17)	129	100 (93–110)	100.23 (13.95)
CELF-4 subtests						
C&FD	32	11 (9.5–12.5)	10.72 (2.47)	129	10 (8–11)	9.19 (2.55)
WS	8	11 (9.5–12.5)	10.25 (3.77)	31	11 (9–12)	10.23 (2.86)
RS	36	11 (8–12)	10.19 (2.35)	129	10 (8–12)	9.84 (3.05)
FS	36	11 (9–12)	10.31 (2.38)	129	10 (9–12)	10.14 (2.54)
WC-R	35	11 (7–12)	10 (2.77)	129	10 (8–12)	9.86 (2.93)
WC-E	35	11 (10–13)	11.23 (2.57)	98	9 (8–12)	9.90 (2.82)
WC-T	35	11 (8–13)	10.69 (2.65)	98	10 (8–12)	10.08 (2.77)

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SS	8	8 (8–12)	8.88 (3.56)	31	10 (7–11)	9.026 (2.54)
WASI/WASI-II						
Non-verbal IQ	36	102.5 (93.0–110.5)	103.64 (11.66)	129	102 (93–112)	102.54 (13.54)

Notes: [a]The identified outlier is excluded.

CELF-4, Clinical Evaluation of Language Fundamentals, Fourth Edition; WASI, Wechsler Abbreviated Scale of Intelligence; WASI-II, WASI, Second Edition; CLP, cleft lip and palate; CP, cleft palate; IQR, interquartile range; SD, standard deviation; CLS, Core Language Score; RLI, Receptive Language Index; ELI, Expressive Language Index; C&FD, Concepts & Following Directions; WS, Word Structure; RS, Recalling Sentences; FS, Formulated Sentences; WC-R, Word Classes—Receptive; WC-E, Word Classes—Expressive; WC-T, Word Classes—Total; SS, Sentence Structure.

Table 3. Difference in CELF-4 language scores between cleft and non-cleft groups, adjusted for non-verbal IQ and birth order[a]

CELF-4 index	Cleft versus non-cleft		
	Coefficient	95% CI	*p*
Core Language Score	3.41	–1.35 to 8.18	0.159
Receptive Language Index	3.82	–0.91 to 8.57	0.113
Expressive Language Index	2.19	–2.70 to 7.07	0.378

Notes: [a]The identified outlier is excluded from the analysis.

CELF-4, Clinical Evaluation of Language Fundamentals—Fourth Edition; CI, confidence interval.

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