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Title:
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Date:
2019-09-01

Citation:
Boyce, J. O., Sanchez, K., Amor, D. J., Reilly, S., Da Costa, A., Kilpatrick, N. & Morgan, A. T. (2019). Exploring the speech and language of individuals with non-syndromic submucous cleft palate: a preliminary report. International Journal of Language and Communication Disorders, 54 (5), pp.767-778. https://doi.org/10.1111/1460-6984.12474.

Persistent Link:
https://hdl.handle.net/11343/286863

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

<RRH>*Communication and submucous cleft palate*

Research Report

Exploring the speech and language of individuals with non-syndromic submucous cleft palate: a preliminary report

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(*Received October 2018; accepted April 2019*)

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1111/1460-6984.12474.

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Abstract

Background: Submucous cleft palate (SMCP) has a heterogeneous presentation and is often identified late or misdiagnosed. Diagnosis is prompted by speech, resonance or feeding symptoms associated with velopharyngeal insufficiency. However, the broader impacts of SMCP on communication have rarely been examined and therefore are poorly understood.

Aims: To describe the communicative profile of individuals with non-syndromic SMCP by examining speech, language and pragmatics (social language).

Methods & Procedures: Fifteen participants with SMCP aged 5;1–12;8, without a genetic diagnosis, participated in the study. Participants completed standardized assessments examining language, resonance, speech and non-verbal intellect. Parents also completed the Children's Communication Checklist (CCC-2), which provided a measure of overall communicative ability, including pragmatic skills. Formal language outcomes were compared with two cohorts: 36 individuals with overt non-syndromic clefts and 129 individuals with no history of clefting.

Outcomes & Results: Speech intelligibility was reduced secondary to hypernasality, disordered articulation and/or impaired phonology (n = 7) in children with SMCP. Poorer overall language outcomes were observed for children with SMCP compared with both those with overt clefts and no history of clefting ($p < 0.001$). Language scores for children with SMCP ranged from impaired (n = 6) to above the standardized mean (n = 4). Receptive and expressive language performance were independently correlated with non-verbal IQ ($p < 0.01$). Those with severe language impairment (n = 4) also had borderline or impaired non-verbal IQ. Parents reported that speech and semantics were the most affected sub-domains of communication, while scores were the highest for initiation domain. Speech and language skills were correlated strongly with pragmatics ($r = 0.877$, $p < 0.01$).

Conclusions & Implications: Overall, performance was variable within the SMCP group across speech, language and pragmatic assessments. In addition to well-documented speech difficulties, children with SMCP may have language or pragmatic impairments, suggesting that further

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neurodevelopmental influences may be at play. As such, for individuals with SMCP, additional clinical screening of language and pragmatic abilities may be required to ensure accurate diagnosis and guide both cleft and non-cleft related therapy programmes.

Keywords: submucous cleft palate, speech, expressive language, receptive language, pragmatics.

<A>**What this paper adds**

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

SMCP has a heterogeneous presentation and is often diagnosed late or misidentified. Identification is typically prompted by speech, resonance or feeding difficulties. Little is known about the broader communication profile of children with SMCP, restricting accurate differential diagnosis and targeted treatment.

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

This study provides the first comprehensive evaluation of the speech, language and pragmatic skills of children with non-syndromic SMCP. Results showed that children with SMCP have diverse communicative profiles, with overall poor language. Those with significant language impairment also had impaired non-verbal IQ, suggesting the coexistence of neurodevelopmental disorders.

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

If children present with SMCP and impaired speech or resonance, treating health professionals should also consider screening broader communication skills, including language and pragmatics. Outcomes can inform comprehensive diagnosis and ensure timely access to appropriate therapy.

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<A>**Introduction**

Submucous cleft palate (SMCP) is a sub-phenotype of overt cleft palate and is most commonly identified through one or more visible intra-oral features: a bifid uvula; a translucent line in the middle of the soft palate (zona pellucida); and a palpable bony notch at the hard and soft palate junction (Calnan 1954). Reported prevalence around the world is 0.02–0.50%; however, these figures are likely an underestimation due to misdiagnosis and under-reporting (Feka *et al.* 2019, Gosain *et al.* 1996, Sales *et al.* 2018, Weatherley-White *et al.* 1972). There is no clear pattern of occurrence across sexes, though a number of studies have reported insignificant differences (Chosack and Eidelman 1978, Feka *et al.* 2019). A diagnosis of SMCP is generally pursued when an individual presents with velopharyngeal insufficiency (VPI) with or without other associated difficulties, including poor feeding skills (Reid *et al.* 2006), middle ear dysfunction (Flynn *et al.* 2013) or hearing loss (Skuladottir *et al.* 2014). Little is known about the broader communication profile of children with SMCP, though language, including receptive, expressive and pragmatic abilities, is an identified area of difficulty for those with overt clefts (Morgan *et al.* 2017, Hardin-Jones and Chapman 2014, Frederickson *et al.* 2006). Individuals with SMCP are extremely diverse in both their anatomical presentation and accompanying symptoms, with many remaining asymptomatic into adulthood (McWilliams 1991). As a result, a correct diagnosis is often delayed or preceded by incorrect diagnoses and treatment, hindering a child's overall development and worsening accompanying difficulties (Ha *et al.* 2013).

<B>*Aetiology*

SMCP can present in isolation or as a phenotypic feature of recognized malformation syndromes and chromosomal aberrations, such as 22q11 deletion syndrome (Leslie and Marazita 2013). Here, we define isolated or 'non-syndromic' as occurring without any identified genetic abnormality. Non-syndromic clefts have elaborate and poorly defined aetiologies, with likely influence from both genetic and environmental factors (Dixon *et al.* 2011). Genetic investigations into non-syndromic SMCP are in their infancy, however preliminary studies suggest that SMCP is a distinct genetic entity, independent of overt cleft phenotypes (i.e., cleft lip, cleft palate or cleft lip and palate; Reiter *et al.* 2015). Comprehensive behavioural phenotyping specific to SMCP is therefore essential to identify more refined traits that may form part of a broader genetic syndrome (Dixon *et al.* 2011).

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<B>*Speech and language*

Most investigations exploring communication in individuals with SMCP focus on the impact of anatomical abnormalities and surgical treatment (Boyce *et al.* 2018a, Swanson *et al.* 2017). Studies consistently report that individuals with SMCP may have VPI characterized by increased nasal resonance (hypernasality), nasal air emission or turbulence and palatalized or glottal articulation (Boyce *et al.* 2018a, Sommerlad *et al.* 2004). Prevalence of cleft-related speech or resonance disorders amongst non-syndromic SMCP cohorts ranges from < 10% to 88%, though figures are expected to be inflated as many studies recruit participants from clinical settings where eligible individuals are more likely to be symptomatic (Kono *et al.* 1981, McWilliams 1991, Ha *et al.* 2013). Existing studies provide useful descriptions of VPI that inform surgical planning and structurally related speech sound therapy. However, they fail to address broader aspects of communication, many of which have been flagged as areas of difficulty in those with overt orofacial clefts (Morris and Ozanne 2003, Hardin-Jones and Chapman 2014, Frederickson *et al.* 2006).

In addition to VPI, individuals with overt clefts may have impaired phonology (i.e., delayed or disordered speech sound processing in language; Klintö *et al.* 2016, Morris and Ozanne 2003), receptive and expressive language (Hardin-Jones and Chapman 2014, Morgan *et al.* 2017, Scherer and D'Antonio 1995) and pragmatic skills (social language; Chapman *et al.* 1998, Frederickson *et al.* 2006, Slifer *et al.* 2004). For the purposes of this study, the term 'language' refers to receptive and/or expressive language abilities, while 'pragmatics' refers to social language abilities. Two recent studies formally examining language suggest that up to 20% of children with overt non-syndromic clefts may experience language impairment, although summative group averages in these studies were comparable between cleft and non-cleft cohorts (Boyce *et al.* 2018b, Morgan *et al.* 2017). Studies exploring pragmatic abilities centre around conversational analyses, and show low conversational assertiveness in 50% and 20% of preschool and school-aged children with cleft lip and palate, respectively (Chapman *et al.* 1998, Frederickson *et al.* 2006). Another study found that during social interactions, children with overt clefts frequently failed to respond to questions and made fewer choices than their non-cleft peers (Slifer *et al.* 2004). While these communicative impairments have been noted in children with overt clefts, no studies have specifically profiled individuals with SMCP.

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The aim of this study was to describe the communicative profile of a cohort of clinically ascertained individuals with non-syndromic SMCP by examining speech, language and pragmatics (social language). Based on the present literature, we hypothesized that children with SMCP would present with either better or worse speech and language than children with overt clefts or no clefts, dependent on the physiological nature of the SMCP and subsequent impact on VPI. Speech–language therapists are often the first health professionals to identify a SMCP (Ha *et al.* 2013) and a greater understanding of the broader communication profile of children with SMCP will inform more targeted clinical assessment and treatment planning.

<A>**Methods**

<B>*Participants*

Participants were recruited between 2017 and 2018 from the Royal Children's Hospital Cleft Registry and clinical database, Melbourne, Australia. They were required to be 5–12 years old at the time of assessment and have a SMCP diagnosed by a plastic surgeon specializing in cleft. This age range was selected as it reflects the primary school years of education in our geographical region of Melbourne. Participants were eligible for inclusion if they: (1) were born with a SMCP only (i.e., no other orofacial cleft such as cleft lip and/or alveolus); (2) had no diagnosed genetic syndrome, additional craniofacial anomalies or known global developmental delay; (3) had no diagnosed moderate to severe intellectual disability; and (4) spoke English as their dominant language. Eligibility was determined from medical files and parent interview on initial contact. A clinical geneticist (D.A.) was consulted to confirm the absence of a diagnosed genetic syndrome. Where clinically indicated, participants had received chromosome microarrays, and all results showed no abnormalities associated with developmental delay or known syndromes. Ethics approval for this study was obtained from the Royal Children's Hospital Human Research Ethics Committee, Melbourne (#27053).

Thirty-nine participants were identified as eligible for inclusion, 24 were contactable and 15 provided written consent to participate (aged 5;1–12;8, mean age = 9;4). Parents declined to participate for the following reasons: too much time away from school for assessment; children feeling anxious about attending hospital appointments; and family circumstances. Comprehensive interviews were systematically conducted by the first named researcher using a standard form that

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covered details of participants' developmental history, demographics and treatment received (e.g., hearing status, speech therapy or surgical procedures). Children's medical files were further reviewed to obtain any missing information.

Two comparison cohorts were identified from a previous study, including individuals with overt non-syndromic cleft palate with or without cleft lip (n = 36, mean age = 10;8) and individuals with no history of clefting (n = 129, mean age = 10;5; Boyce *et al.* 2018b, Reilly *et al.* 2018). No comparison participants had syndromic diagnoses or co-occurring medical conditions likely to impact their speech or language development, such as epilepsy or traumatic brain injury. They too all spoke English as their dominant language. There was no significant difference in age between SMCP and comparison groups ($p < 0.05$). Maternal education levels ranged from < 12 years of schooling to a university degree. Formal language and non-verbal IQ data were available for these cohorts, both assessed using the measures outlined below. Language and non-verbal IQ scores for the overt cleft and non-cleft groups fell within the assessment standardized normative ranges; mean core language scores (CLSs) = 103 (standard deviation (SD) = 10.31) and 99.13 (SD = 13.72) and non-verbal IQ = 103.64 (SD = 11.66) and 102.54 (SD = 13.54), respectively.

<B>*Speech and resonance measures*

Speech and resonance were examined using a common cleft speech assessment protocol, which consists of automatic speech (i.e., counting), a conversational speech sample and a set of sentences examining all English consonants and clusters in relevant word positions (Henningsson *et al.* 2008, Sell *et al.* 1999). Sentence stimuli were obtained from the Great Ormond Street Speech Assessment (GOS.SP.ASS; Sell *et al.* 1999). Intelligibility was rated on a four-point severity scale, with 0 indicating that speech was always easy to understand and 3 indicating severely unintelligible speech (Henningsson *et al.* 2008). Hypernasality was rated on a similar four-point scale, where 0 = normal resonance and 1–3 = mild, moderate or severe hypernasality (Sell *et al.* 1999). Listeners noted whether nasal air emission and turbulence were present or absent, and if present, the number of sounds affected. An oral examination was completed by the primary examiner to identify any structural anomalies that may have impacted on speech. Where phonological impairment was suspected from cleft speech assessment, the Phonology subtest of the Diagnostic Evaluation of Articulation and Phonology (DEAP) was also completed (Dodd *et al.* 2002). The Phonology subtest

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includes 50 single words designed to elicit a range of speech sounds and combinations for subsequent analysis of phonological processes.

All speech samples were rated live by a trained speech–language therapist (J.B.) and recorded with an HD camera connected to a D55S AKG acoustic dynamic microphone for reliability testing. A perceptual reliability study was completed for all participants with SMCP. Audio-video recordings were re-rated once by the primary assessor (J.B.) and twice by an independent blinded speech–language therapist (K.S.) to determine intra- and interrater reliability. Both assessors had over 5 years of postgraduate clinical experience in perceptual speech assessment. Reliability was determined using percentage agreement and weighted kappa coefficients across all perceptual measures. Interrater analysis was completed using the first ratings made by both listeners.

A quantitative measure of resonance was obtained from the Nasometer II, 6450 model (KayPENTAX). Participants aged 11 years and above recorded two standardized reading passages: the Zoo passage, 0% nasal phonemes (Fletcher 1972); and the Rainbow passage, 11.5% nasal phonemes (Fairbanks 1960). Younger participants (i.e., aged $\leq$ 10 years) recorded four sets of sentences from the Simplified Nasometric Assessment Procedures, each targeting a different group of consonants: bilabial, alveolar, velar and sibilant (Kummer 2005, KayPENTAX 2010). The Nasometer was calibrated before each recording session and fitted according to the manufacturer's instructions (KayPENTAX 2010). Recorded samples were examined and edited to exclude any extraneous elements (e.g., recording errors, laughing or loud background noises) and nasalance scores were extracted using the Nasometer II computer software. Nasometer test–retest reliability results are reported in another study that used the same methods and Nasometer machine (Boyce *et al.* 2018c). Recordings were repeated twice within one session for 49 participants (n = 32 recorded the passage stimuli and n = 17 recorded the sentence stimuli). Results showed strong correlation between recordings (Pearson's $r < 0.800$, $p < 0.01$) across all nasalance stimuli, indicating good test–retest reliability (Portney and Watkins 2000).

Nasalance scores were compared with normative paediatric Australian data reported by Boyce *et al.* (2018c) as follows (mean (SD)): Zoo passage = 10.5 (4.8); Rainbow passage = 32.6 (5.8); bilabial sentences = 9.5 (3.3); alveolar sentences = 10.3 (4.3); sibilant sentences = 13.4 (5.2); and velar sentences = 11.0 (4.2). A score $\geq$ 2 SD above the normative mean was selected as the clinically significant threshold value for each stimulus (Kummer 2005, KayPENTAX 2010).

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<B>*Language and pragmatics*

Receptive and expressive language skills were assessed using the Australian Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4), a valid and reliable standardized tool (Semel *et al.* 2003). The CELF-4 generates three language index scores: CLS reflecting overall receptive and expressive abilities, receptive language index (RLI) and expressive language index (ELI). All index scores have a normative standardized mean of 100 and SD of 15. Individual subtests assess aspects of syntax, morphology and semantics with a standardized average of 10 (SD = 3). In this study, language impairment was defined as a standard score ≥ 1.25 SD below the standardized mean (McKean *et al.* 2017, Reilly *et al.* 2018).

A measure of non-verbal intellect was obtained from the Wechsler Abbreviated Scale of Intelligence, 2nd edn (WASI-II; Wechsler 2011). Block design and matrix reasoning subtests were completed to generate a summative measure, the performance reasoning index (mean = 100, SD = 15). Non-verbal IQ was considered alongside language to account for any interactions that may be occurring (Gallinat and Spaulding 2014).

Parents completed the Children's Communication Checklist, 2nd edn (CCC-2), which provided a measure of each participant's overall communicative ability (Bishop 2003, Norbury *et al.* 2004). The CCC-2 uses parents' observations of their children across contexts, providing a representative depiction of each child's abilities (Norbury *et al.* 2004). The 70-question checklist generates scales that cover speech, language (syntax, semantics and coherence), pragmatics and social skills. The pragmatics scale includes initiation, stereotyped language, use of context and non-verbal communicative ability. Australian standardized mean scaled scores range from 8.63 to 9.53 (SD = 2.85–3.34). A general communication composite is obtained from the sum of all scales and a score < 55 indicates communication impairment. For those with scores < 55, a social interaction deviance composite is also generated by comparing the speech and language results with those reflecting social interaction abilities (initiation plus non-verbal communication plus social relations plus interests).

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<B>*Data analysis*

Speech, language and pragmatic outcomes were determined for each participant with SMCP. The associations between (1) receptive and expressive language domains (CELF-4) versus non-verbal IQ (WASI-II); and (2) speech and language (CCC-2) versus pragmatics (CCC-2) were quantified using Pearson's correlation coefficient (*r*). To account for multiple comparisons, a Bonferroni-adjusted significance threshold of $p < 0.01$ was applied.

Core language outcomes from the CELF-4 were also compared with results from those with overt clefts and no history of clefting, through descriptive analysis and multivariate linear regression. Maternal education level (a factor known to be important in language development), age and non-verbal IQ were controlled for in the analysis (McKean *et al.* 2017).

<A>**Results**

Age at SMCP diagnosis ranged from birth to 5 years, with most diagnoses prompted by speech, resonance or feeding difficulties (see table 1 for demographic data). Thirteen participants had undergone primary surgical repair of their SMCP before taking part in this study. Of these 13, two participants had also undergone further surgery to reduce VPI (participants 13 and 14; table 1) and one was on the waiting list for a secondary procedure (participant 3). The remaining two participants had unrepaired SMCPs as their clefts had not given rise to clinically significant symptoms, as judged by their treating cleft teams. Twelve participants had a history of otitis media or ear infections, and nine participants had received grommets in the past. All but one (participant 7) had received at least 6 months of speech therapy before assessment for this study.

<tab 1>

A range of speech and resonance profiles were found across the cohort, confirmed by perceptual and instrumental measures (table 2 and figure 1). Reduced intelligibility was noted in seven participants secondary to a combination of hypernasality, disordered articulation and/or impaired phonology. Across the group, hypernasality ranged from normal to moderate; and nasal air emission or turbulence ranged from absent to apparent on more than three consonants. The most common articulatory and phonological errors were lateralization of anterior sounds ($n = 4$) and gliding of /r/ ($n = 4$; see table S1 for detailed speech error profiles). On oral examination, one

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participant had a class III malocclusion (number 15). No participants had tongue ties, palatal fistulae or any other structural challenges that may have impacted on speech, such as missing teeth. An equivalent range in outcomes was reflected in nasalance scores (figure 1), where a similar number of participants scored above or below the clinically significant threshold for one or more speech stimuli.

Agreement in perceptual ratings within and between listeners was high (> 86%) across all parameters. Kappa coefficients for interrater reliability ranged from substantial (k = 0.61–0.80) to perfect (k = 1.00, phonology ratings; Landis and Koch 1977). Both listeners 1 and 2 had substantial to perfect intra-rater agreement across all parameters apart from hypernasality and nasal air emission, which had poor to moderate agreement (listener 2 only). Perceptual speech reliability results are shown in table S2.

<tab 2, fig 1>

Language profiles also varied across the SMCP group (figure 2). Six participants (40%) had language impairment (CLS ≥ 1.25 SD below the standardized mean), and language was severely impaired in four of these six (> 2 SD below the standardized mean). These four participants also had impaired non-verbal IQ. By contrast, four participants achieved overall language scores above the standardized mean. Across language sub-domains, participants achieved the lowest scores in the Word Structure subtest, which examined morphology, and highest scores in the Word Classes – Expressive subtest, examining expressive semantic abilities (see table S3 for performance on individual subtests). Amongst those with impaired language, two also had mild hypernasal resonance, three had accompanying nasal air emission or turbulence, one had disordered articulation and five had delayed and/or disordered phonology. Across all participants with SMCP, both receptive and expressive language domains were strongly correlated with non-verbal IQ; r = 0.733 and 0.757, respectively ($p < 0.01$).

<fig 2>

Figure 3 shows the number of children with receptive and expressive language impairment across SMCP, overt cleft and non-cleft groups. Children with SMCP achieved significantly lower CLSs than those with overt clefts (coefficient = 17.91, confidence interval (CI) = 8.92–26.90, $p < 0.001$) and those with no history of clefting (coefficient = 14.91, CI = 7.02–22.81, $p < 0.001$). A further analysis was also conducted, removing the four individuals in the SMCP group who could be considered as

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outliers (i.e., those with significantly impaired language and non-verbal intellect; figure 2). The findings showing the SMCP group had poorer language than the overt cleft group held (coefficient = 10.44, CI = 1.64–19.23, $p < 0.05$), but differences became insignificant between those with SMCP and no history of clefting (coefficient = 7.06, CI = –0.83 to 14.96, $p = 0.079$).

<fig 3>

Results from parent reported outcomes (CCC-2) also varied across the sample. Median and interquartile ranges of scaled scores for each domain of the CCC-2 are shown in figure 4. The lowest median scores were seen in the speech and language (semantics) sub-domains. Performance was best on the sub-domains of initiation, coherence and social skills. Overall, the speech and language summative score was positively correlated with pragmatics, $r = 0.877$ ($p < 0.01$). Four participants (27%) achieved < 1 SD below the standardized mean on one or more of the four pragmatic categories. Six participants achieved a general communication composite < 55 indicating language impairment. Of these participants, five obtained a social interaction deviance composite < 0, suggesting that their difficulties were predominantly with speech and language rather than social skills. One individual (participant 10) achieved a score of –1, indicating that their pragmatic and social difficulties may be disproportionate to their language impairment, although marginally.

<fig 4>

<A>**Discussion**

Here we provide the first description of speech, language and pragmatic abilities of a cohort of children with SMCP. Communicative profiles varied significantly across the group. Receptive and expressive language scores were correlated with non-verbal IQ and ranged from severely delayed to above the normative average range. A similar range was found in speech and pragmatic presentations. The broad spectrum of results across all communicative parameters suggests that beyond structural anomalies, underlying developmental influences may also be at play.

Speech profiles reported in this study complement existing literature, with participants having normal to severe hypernasality accompanied by a range of cleft speech articulatory features (Boyce *et al.* 2018a, Isotalo *et al.* 2007). Almost half the participant group ($n = 7$) had speech that was mildly to moderately reduced in intelligibility and characterized by hypernasality, increased

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nasalance, disordered articulation and/or impaired phonology, while the remaining participants had speech that was always 'easy to understand'. Further, a high proportion (53%) of participants had delayed phonology on at least one speech sound. The heterogeneity of speech profiles reported here may be anticipated given the broad age range of participants and age at SMCP diagnosis, with further associated influences of individualized surgical treatment (e.g., timing and technique; Boyce *et al.* 2018a, Swanson *et al.* 2017, Sommerlad *et al.* 2004). The wide range of age at diagnosis (0;0–5;1) and surgical treatment (0;10–8;5) in our small cohort is suggestive of a remarkable variety seen in this subtle yet distinct anomaly.

To our knowledge, this is the first study to explore the language and pragmatic skills of children with SMCP alongside speech, limiting possible comparisons to existing literature (Boyce *et al.* 2018a). Nonetheless, we can draw inferences from studies that examine these skills in children with overt clefts. Studies using the same standardized language assessment (CELF-4) show similar findings, with language outcomes ranging from severely impaired to above the average range (Boyce *et al.* 2018b, Morgan *et al.* 2017). However, 40% of our participants with SMCP had impaired language, double the largest proportion reported amongst overt cleft groups (Morgan *et al.* 2017). It is of note that the four participants with SMCP and severely impaired language were the only individuals to also have borderline or impaired non-verbal IQ. It may be hypothesized that these four participants have additional neurodevelopmental disorders not yet identifiable from available clinical or genetic investigations.

Language comparisons with overt cleft and non-cleft groups using CELF-4 scores revealed high coefficients, showing that as a group, those with SMCP had significantly poorer language performance. It was possible, however, that the SMCP group results were negatively skewed because of the four participants with severe language impairment. When these participants were removed, differences held between SMCP and overt cleft groups but not between the SMCP and non-cleft groups. Nonetheless, for the latter comparison, there was a high positive coefficient and wide confidence interval above zero, suggesting that the lack of significance here may be secondary to small sample size. Therefore, further investigations of similar larger cohorts are required to generate stronger conclusions regarding the overall language capabilities of children born with SMCP.

From a parental perspective, findings were similar to the clinical speech and language outcomes, with diverse patterns in overall communication across all sub-domains of the parent

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questionnaire (CCC-2). Speech was the lowest scoring subtest, as expected, given that SMCP is commonly associated with VPI and subsequent articulation and resonance difficulties. In relation to pragmatic skills, existing studies examine different domains (i.e., assertiveness, responsiveness and social behaviours) to the CCC-2, limiting direct comparison with results from this study (Slifer *et al.* 2004, Chapman *et al.* 1998, Frederickson *et al.* 2006). Nevertheless, the proportion of children with impairment in one or more pragmatic scale in this study (27%) falls within the range of those with low assertiveness from previous studies (20–50%; Chapman *et al.* 1998, Frederickson *et al.* 2006). Our results suggest an association between speech and language skills and pragmatic abilities, highlighting pragmatics as an additional area for investigation and potential treatment amongst this population. Further exploration of the precise relationship between speech, language and pragmatics in comparison with other cohorts (e.g., overt cleft or non-cleft) is warranted.

<B>*Strengths and limitations*

The detailed protocol from this study enabled phenotyping of speech, language and pragmatic skills of children with SMCP. Qualitative perceptual speech data were supported by quantitative nasalance measures that showed similar results. The cross-sectional study design and specific research protocol meant that recruited participants spanned a large age range and had received intervention, both surgical and therapeutic, at different ages. Further, most participants had a repaired SMCP at the time of assessment. This restricted the data available on pre-surgical anatomical presentation and introduced a significant confounding factor over which we had no control. Given the heterogeneity across our sample, it was not possible to account for SMCP subtype, nor the methods of surgical and therapeutic interventions.

It is also important to note that the SMCP cohort for this study was recruited from a clinical database, introducing a potential selection bias. In contrast to overt clefts, if a SMCP is asymptomatic, its subtle nature means that referral and diagnosis may be delayed, or not occur (Ha *et al.* 2013). Therefore, results from this preliminary study highlight outcomes from a clinically ascertained population, which may not be an accurate representation of the population of individuals with SMCP as a whole. Nonetheless, the significant range in communication outcomes beyond structural impacts (i.e., high proportions with impaired language and varied pragmatic abilities), highlights a number of observations worthy of further exploration. In a larger longitudinal

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cohort, additional relationships could be explored, such as differences between speech and language presentation, the impact of fluctuating hearing status, or the effect of surgical and therapeutic interventions on communicative outcomes.

<B>*Clinical implications*

We found that some individuals with isolated SMCP may present with additional difficulties beyond those directly associated with their anatomical anomaly. This knowledge is relevant for the multidisciplinary clinical treating team and may lead to further investigations or referrals to examine underlying or associated aetiologies. Results are particularly pertinent for speech–language therapists, as many SMCP diagnoses are prompted by speech difficulties. If a child has an identified SMCP and associated speech difficulties, it may be beneficial to also screen their broader communication skills, including language and pragmatics, alongside cognition. The CCC-2 could be a useful tool for this screening purpose, as it provides standardized information across various communication domains. The CCC-2 is easy to use and can be completed by carers in the clinic waiting room. Presenting symptoms can then be considered as part of the child's overall presentation to inform both cleft and non-cleft-related treatment programmes.

<A>**Conclusions**

This exploratory paper highlights heterogeneity in the communication profiles of children born with SMCP without an identified genetic syndrome. Speech, language and pragmatic skills ranged from above the average range to severely impaired. In this clinically ascertained cohort, children with SMCP achieved poorer language outcomes than those with overt clefts or no history of clefting. Individuals with the most severe language impairment were also the only participants with SMCP to have impaired non-verbal IQ, suggesting a possible influence from additional developmental factors. As such, if an individual presents with SMCP and poor speech, clinical assessment and treatment plans should also consider broader communication profiles, beyond features associated with anatomical presentation.

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<A>**Acknowledgements**

The authors thank the participants and their families for their involvement in this study. This work was supported by a National Health and Medical Research Council (NHMRC) Practitioner Fellowship #1105008 and Centre of Research Excellence (CRE) in Speech and Language (SLANG) #1116976 awarded to A.M., a NHMRC-funded project, OzCleft (#607396) awarded to N.K., and the Murdoch Children's Research Institute Postgraduate Health Research Scholarship awarded to J.B. This work was also supported by the Victorian government's Operational Infrastructure Support Programme. ***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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Table 1. Participant characteristics.

Participant	Sex	Age at assessment	Age at diagnosis	Reason for the diagnosis	Age at primary repair	Maternal education	Family history of cleft?	Hearing status at the assessment[b]	Non-verbal IQ[c]
1	F	12;6[a]	4;4	Speech/resonance difficulties (VPI)	n.a.	Tertiary	–	WNL	–2 SD
2	M	12;8	4;6	Speech/resonance difficulties (VPI)	8;5	Tertiary	–	WNL	WNL
3	M	10;1	4;6	Speech/resonance difficulties (VPI)	4;11	< 12 years[d]	–	WNL	WNL
4	F	11;9	1;6	Speech/resonance difficulties (VPI)	2;3	Tertiary	+	WNL	WNL
5	M	12;6	n.r.	n.r.	8;0	< 12 years[d]	–	WNL in left, mild conducti	WNL

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								ve loss in right	
6	F	9;10	5;1	Speech/resonance difficulties (VPI)	5;9	Tertiary	+	WNL	WNL
7	F	7;9	0;6	Feeding difficulties	1;1	Tertiary	+	WNL	WNL
8	M	5;6	0;2	n.r.	0;10	Tertiary	+	Mild conductive loss	WNL[e]
9	F	6;11	0;10	Oral examination	2;11	Training certificate	–	WNL	WNL
10	F	8;5	0;1	Feeding difficulties	0;8	12–13 years[d]	–	WNL	WNL
11	F	10;6	0;1	Feeding difficulties	0;8	Tertiary	+	WNL	WNL
12	M	7;5	3;11	Speech/resonance difficulties (VPI)	5;8	Tertiary	+	WNL	–1 SD
13	M	8;10	3;8	Speech/resonance	3;11	Tertiary	–	WNL	–2 SD

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				difficulties (VPI)					
14	F	9;10	0;0	Feeding difficulties	0;10	12–13 years[d]	+	WNL	WNL
15	F	5;1	4;11	Oral examination	n.a.	12–13 years[d]	–	WNL	–1 SD[e]

Notes: [a]All ages are reported as year;months.

[b]Normal hearing was classified as ≤ 20 dB loss across frequencies, as determined from parent interviews and/or reference to medical files.

[c]Perceptual Reasoning Index, Wechsler Abbreviated Scale of Intelligence, Second edition.

[d]Years of education.

[e]Standardized score is considered as descriptive only as participants were younger than the lowest standardized age of the test. Reported information was scored using the youngest available standardized data (6;0 years).

M, male; F, female; VPI, velopharyngeal insufficiency; n.r., not reported; n.a., not applicable; WNL, within normal limits; SD, standard deviation.

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Table 2. Perceptual speech results

Participant	Intelligibility	Hypernasality (consistent)	NAE/NT	Articulation	Phonology
1[a]	Mild	WNL	WNL	WNL	Delayed
2	Mild	Mild	WNL	WNL	Delayed
3	Mild	Moderate	NAE NT	Disordered	WNL
4[a]	Mild	WNL	WNL	Disordered	Delayed
5	WNL	Mild	WNL	WNL	WNL
6	WNL	WNL	WNL	WNL	WNL
7	WNL	WNL	WNL	Disordered	WNL
8	WNL	WNL	NT	WNL	WNL
9	WNL	WNL	WNL	WNL	Delayed
10	WNL	WNL	NT	WNL	Delayed
11	WNL	WNL	WNL	WNL	WNL
12	Moderate	WNL	NAE	Disordered	Delayed + disordered

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13	Mild	Mild	NT	WNL	Delayed
14	WNL	WNL	NT	WNL	WNL
15[b]	Moderate	WNL	WNL	WNL	Delayed

Notes: [a]These participants also had inconsistent general imprecision and phonetic distortions in connected speech.

[b]This participant also had a number of inconsistent disordered speech patterns, for example, word medial intrusive consonant.

NAE/NT, audible nasal air emission/nasal turbulence; WNL, within normal limits. A shaded box = atypical feature (darker = more severe).

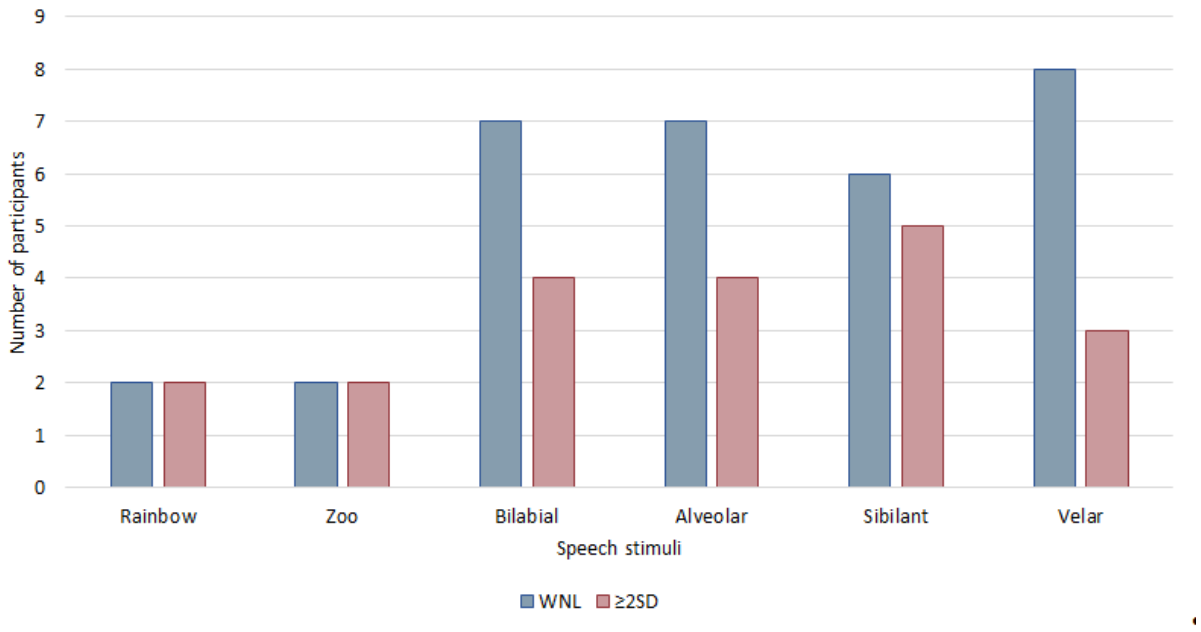


Figure 1. Number of participants achieving typical (< 2 SD) versus clinically high (≥ 2 SD) nasalance scores across speech samples. SD, standard deviation.

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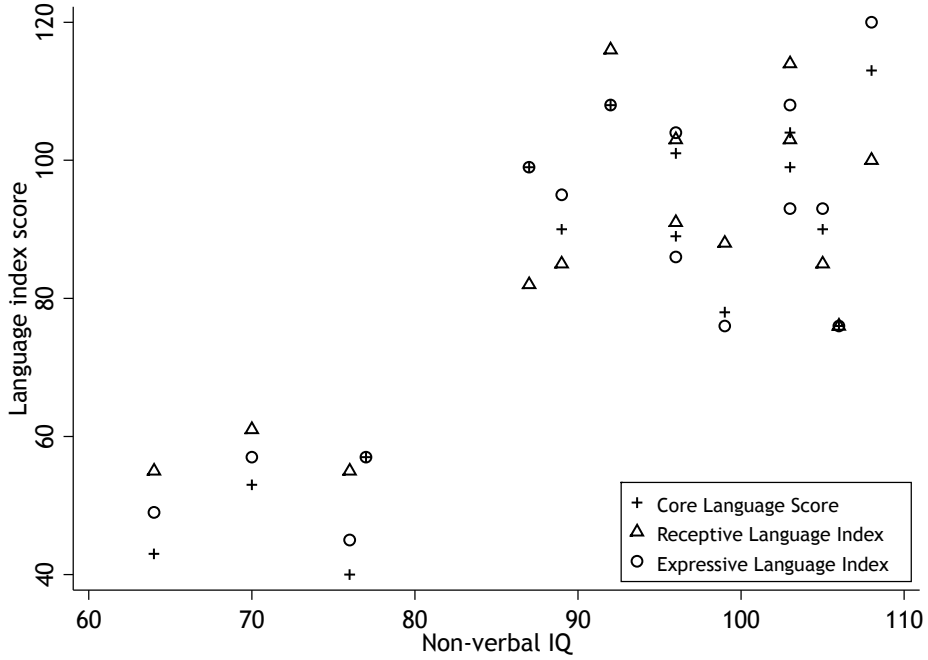


Figure 2. Language index scores against non-verbal IQ for participants with submucous cleft palate (SMCP).

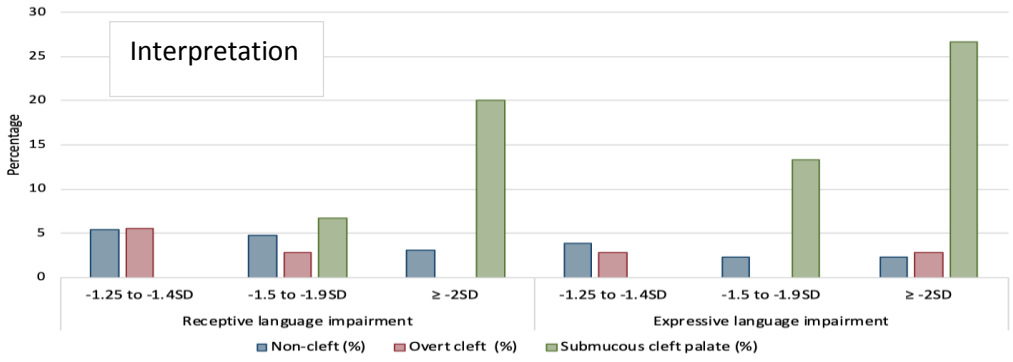


Figure 3. Participants with language impairment (%). Both receptive and expressive language impairment was found for 27% (*n* = 4) of children with SMCP and 4% (*n* = 5) of children with no history of clefting. SD, standard deviation.

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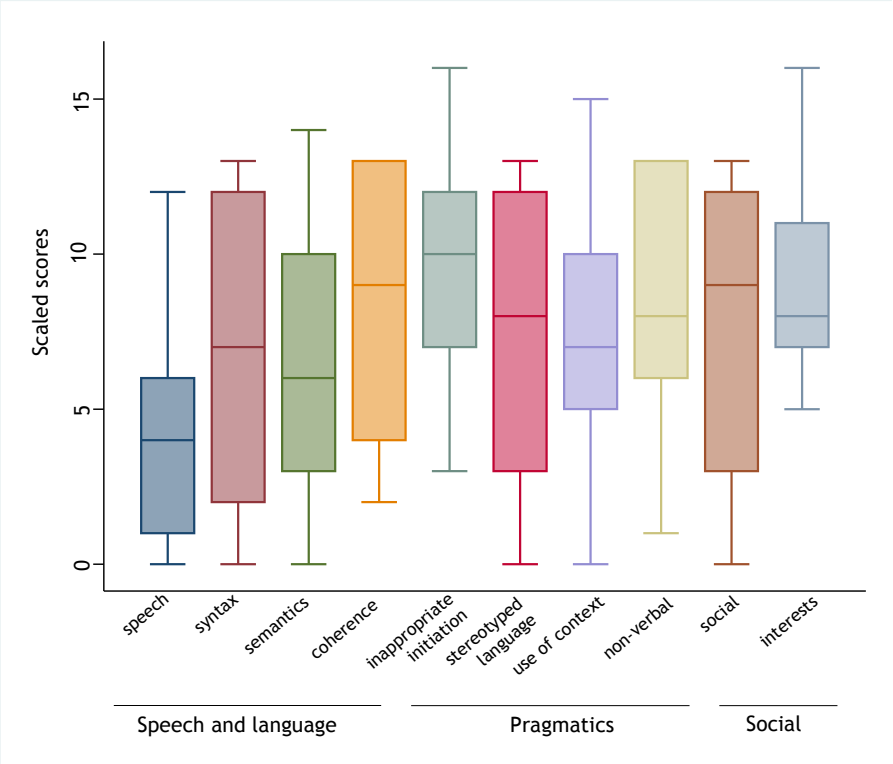


Figure 4. Speech and language, pragmatics and social profiles from the parent questionnaire (Children's Communication Checklist, 2nd edn).

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