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**Descriptive Title:** Speech and language characteristics in individuals with non-syndromic submucous cleft palate – a systematic review

**Short Title:** Non-syndromic submucous cleft palate speech

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## **Abstract**

### **Background**

Up to 80% of individuals with unrepaired submucous cleft palate (SMCP) experience speech difficulties secondary to velopharyngeal insufficiency. Language delays are reported in the broader cleft lip and/or palate population, suggesting that individuals with SMCP may also be at risk. However contemporary understanding of this population remains limited as there has been no systematic examination of the literature. This review aims to systematically review and document the speech and language characteristics of individuals with non-syndromic SMCP. In addition, to identify factors reported to impact speech and language outcomes.

### **Method**

This review followed the PRISMA guidelines. Five databases were comprehensively searched using keywords and indexed headings. Included studies had to report speech or

language outcomes of individuals with non-syndromic SMCP. Risk of bias and methodological design quality were examined using tools from the Scottish Intercollegiate Guidelines Network. Relevant data was extracted for analysis.

### **Results**

Eighteen studies met inclusion criteria, yielding 598 participants. Study results showed that individuals with unrepaired non-syndromic SMCP may have speech difficulties secondary to velopharyngeal insufficiency including increased nasal resonance and palatalised or glottal articulation. Primary surgical repair between three and four years of age led to better post-surgical speech outcomes. There is a paucity of literature outlining motor or phonological aspects of speech and receptive or expressive language abilities of this population.

### **Conclusion**

Individuals with non-syndromic SMCP present with speech difficulties similar to those experienced by individuals with overt cleft palate. Health care professionals should be aware of possible presenting symptoms and consider early SMCP diagnoses where appropriate. Further research is needed to specify the broader communication profile in this population.

## Introduction

Submucous cleft palate (SMCP) is a distinct sub-phenotype of cleft palate, characterised by palatal muscle defects with intact oral surface mucosa (Calnan, 1954; Sommerlad *et al.*, 2004; Velasco *et al.*, 1988). An overt SMCP is visible intra-orally and identified from one or more of Calnan's triad: a bifid uvula, zona pellucida and palatal notch (Calnan, 1954; Kelly, 1910). An occult SMCP describes underlying velar malformation without classic intra-oral findings, and can only be detected through direct visualisation of the superior palatal surface by nasoendoscopy or surgical dissection (Gosain *et al.*, 1996; Kaplan, 1975). Prevalence of SMCP amongst children is reported at 0.02-0.08% (Kono *et al.*, 1981; Shprintzen *et al.*, 1985; Velasco *et al.*, 1988; Weatherley-White *et al.*, 1972). However, precise tracking is difficult as diagnosis and treatment are often undesirably delayed into mid-childhood and even adulthood (Reiter *et al.*, 2011; Weatherley-White *et al.*, 1972). Here we define non-syndromic SMCP as occurring independent of commonly identified genetic syndromes such as velocardiofacial syndrome.

In addition to structural abnormalities, individuals with SMCP may also experience difficulties with feeding, middle ear function and hearing. Impaired palatal muscles can prevent a baby from generating negative intra-oral pressure required for feeding; leading to lengthened and difficult feeds accompanied by possible nasal regurgitation (Bessell *et al.*, 2011; Ha *et al.*, 2013). These challenges are overcome with specific feeding techniques and bottles (Bessell *et al.*, 2011). Soft palate defects also restrict eustachian tube dilation and opening, resulting in otitis media with effusion and associated hearing loss (Flynn *et al.*, 2013). Despite this array of symptoms, a diagnosis of SMCP is generally only pursued when an individual presents with persistent velopharyngeal insufficiency (VPI) and disordered speech (Oji *et al.*, 2013; Park *et al.*, 2000; Velasco *et al.*, 1988). VPI is detected when a child begins to speak and symptoms may be extremely subtle (Ha *et al.*, 2013; Reiter *et al.*, 2011; Sullivan *et al.*, 2011).

Speech difficulties associated with VPI occur in up to 80% of individuals with unrepaired SMCP (Ha *et al.*, 2013; Kono *et al.*, 1981; Shprintzen *et al.*, 1985). Defective palatal muscles prevent adequate velopharyngeal closure during speech, allowing undesired

air and acoustic energy to escape into the nasal cavity. Resulting speech is characterised by increased nasal resonance (hypernasality), nasal air emissions (NAE) or turbulence and passive articulation characteristics (e.g., weak or nasalised consonants). These structurally based features are persistent and contrast mis-learned oral and non-oral articulatory errors also common amongst individuals with clefts (John *et al.*, 2006). Oral errors are classified based on their placement within the oral cavity and identified as anterior (e.g., lateral or palatal) or posterior (e.g., backed to velar or uvular). Non-oral errors include pharyngeal and glottal articulation along with active nasal fricatives (Sell *et al.*, 1999). Across literature in the field, cleft speech passive and mis-learned patterns are interchangeably described as obligatory distortions (i.e., secondary to structural abnormalities) and compensatory errors (i.e., changes in place of articulation to counter structural limitations) respectively (Baek *et al.*, 2017; Isotalo *et al.*, 2007; Kummer, 2011; Ng *et al.*, 2015). Differential diagnosis of presenting speech symptoms – i.e., structural versus mis-learned errors – can be challenging and is often facilitated by diagnostic speech therapy to inform treatment decision making (Marsh, 2004). Further to articulation difficulties, increased phonological errors are reported in cleft palate populations, but not routinely considered amongst individuals with SMCP (Chacon *et al.*, 2017; Chapman, 1993; Schönweiler *et al.*, 1999). Phonological error patterns occur at a cognitive-linguistic level and manifest in errors of speech sound processing rather than consonant production (Dodd *et al.*, 1989; Grunwell, 1975).

Broader communication profiles considering language and cognitive skills are not widely documented amongst the SMCP population. However, difficulties in expressive and receptive language (Broen *et al.*, 1998; Hardin-Jones *et al.*, 2014; Scherer *et al.*, 1995), literacy (Gallagher *et al.*, 2017; Lee *et al.*, 2015; Wehby *et al.*, 2014), and cognition (Nopoulos *et al.*, 2002; Roberts *et al.*, 2012) are consistently reported amongst the broader cleft lip and/or palate group, suggesting that individuals with SMCP may also be at risk.

Speech and language outcomes are complicated by a multitude of associated factors including hearing abilities (Moeller, 2000; Yoshinaga-Itano *et al.*, 1998), socioeconomic status (Calvo *et al.*, 2014; Hoff, 2003), social exposures (Hoff, 2006; Tamis-LeMonda *et al.*, 2004) and bilingualism (Calvo *et al.*, 2014; Costa *et al.*, 2004; Hambly *et al.*, 2013).

Additional confounding factors specific to SMCP include age at diagnosis, anatomical features, access to speech therapy and surgical correction (Bezuhly *et al.*, 2012; Ha *et al.*, 2013; Schönweiler *et al.*, 1999; Sommerlad *et al.*, 2004). Speech therapy is required to treat habitual oral and non-oral consonant production errors and can thus occur at any time (i.e., before or after surgery). Contrastingly, surgical intervention is indicated when speech difficulties are structurally related and therefore not responsive to therapy. Surgical intervention alters the structure and function of the palate and/or velopharyngeal port to facilitate adequate closure for speech. However, the optimal timing of surgery and suitable operative technique remains controversial (Gilleard *et al.*, 2014; Oji *et al.*, 2013; Sullivan *et al.*, 2011).

To date there has been no systematic examination of speech and language characteristics associated with SMCP. Further, there has been no review examining potential factors that influence these communication outcomes. For both clinicians and researchers, robust and clearly defined clinical characteristics can inform timely and accurate SMCP diagnosis and treatment. Results from this study can also elucidate speech and/or language features associated with SMCP to inform new investigations of both syndromic and non-syndromic cases. The purpose of this study is: (1) To systematically review and document the speech and language characteristics of individuals with un-repaired non-syndromic SMCP; (2) To identify and describe factors reported to impact speech and language outcomes amongst this population.

## Methods

This review follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher *et al.*, 2009).

### **Inclusion and exclusion criteria determining eligibility**

Participants: Studies of non-syndromic participants with overt or occult SMCP only (i.e. no accompanying cleft lip, cleft palate or other anatomical anomalies) were included. Participants were denoted as non-syndromic if they satisfied one or more of the following: (1) were labelled as non-syndromic; or (2) did not present with: additional craniofacial

malformations or deformities; a syndromic diagnosis; intellectual disability; or severe neurological deficits. To encompass all available data, studies of non-syndromic SMCP alongside syndromic SMCP or other orofacial clefts (e.g., cleft lip or palate) were included provided that demographic and outcome data could be extracted for our group of interest. Due to inconsistencies throughout the literature, there were no restrictions on the criteria or method of SMCP diagnosis (i.e., approaches to diagnosis across studies required varying clinical anatomical markers – from one to all of Calnan’s classic triad – identified intra-orally or through nasoendoscopy).

Outcome measures: Studies reporting either quantitative (i.e., standardised test scores or acoustic measures) or qualitative (i.e., perceptual speech ratings) speech or language outcomes were included. Outcomes reported either before or after treatment (i.e., surgery or speech therapy) were accepted with intervention noted clearly.

Study type: We required studies to have at least five participants with non-syndromic SMCP. All study types and levels of evidence published in English were included. No exclusion criteria were set for age and sex of participants.

#### **Search strategy and selection of studies**

A systematic search was conducted using five databases from their inception, until July 2018: MEDLINE (Ovid); EMBASE (Ovid); Cochrane Library; CINAHL (EBSCO); and PubMed. The first four databases were searched using relevant medical subject heading (MeSH) terms and keywords. PubMed was searched using keywords only to retrieve relevant e-pubs and items not indexed in Medline. Key search terms included: terms referring to submucous cleft palate, including the broader cleft palate; and terms relating to VPI, communication, speech and language. Complete search strategies for each database are outlined in Table SI (online supporting information). Individual searches of key journals and publications by experts in the field were also conducted.

All identified references were exported to EndNote X7.7 and duplicates were removed (Thomson Reuters, 2016). Titles, keywords and abstracts were screened by JB to ensure that papers (1) discussed humans diagnosed with SMCP; (2) reported speech or

language outcomes; and (3) were published in English. After screening, full articles were obtained and evaluated for eligibility by JB, in collaboration with AM and NK where necessary. Articles that did not meet the pre-determined inclusion criteria were excluded.

### **Data collection**

Data were extracted from included studies and tabulated by JB and AM. Extracted study characteristics include: study type; number of eligible participants; mean age at diagnosis; presence and type of comparison group; SMCP anatomical features; pre-diagnosis symptoms and treatment; and study exclusion criteria. Details of treatment and outcome measures were also extracted. Outcome data included: hearing status; perceptual and instrumental (e.g., nasometry) speech measures; language abilities; and instrumental measures of velopharyngeal functioning (e.g., nasoendoscopy and videofluoroscopy). Finally, reviewers noted factors reported to impact speech and language outcomes in respective studies; e.g., surgery type (Afrooz *et al.*, 2015; Calis *et al.*, 2018; Ezzat *et al.*, 2016; Park *et al.*, 2000), age at surgery (Baek *et al.*, 2017; Bezuhly *et al.*, 2012; Oji *et al.*, 2013; Swanson *et al.*, 2017) and pre-operative speech characteristics (Baek *et al.*, 2017; Bezuhly *et al.*, 2012).

Two different quality assessment tools were trialled for this review, namely the Newcastle-Ottawa Scale (Wells *et al.*, 2005) and the Scottish Intercollegiate Guidelines Network (Harbour *et al.*, 2001). The latter was chosen for its descriptive checklists and close implementation of the widely-adopted core GRADE principles (GRADE, 2004). Strength of evidence was first determined by identifying a level of evidence using the SIGN algorithm (Scottish Intercollegiate Guidelines Network (SIGN), cited 2017). Levels of evidence were assigned as 1 (randomised controlled trials), 2 (case-control or cohort studies) or 3 (case series). Studies were then assigned a quality rating (-, + or ++) based on the selection of participants, assessment reliability and validity, identification of confounding factors and statistical analysis (Harbour *et al.*, 2001). Differences in subject groups and confounders were considered with more weighting in studies where factors impacting speech outcomes were discussed. To include all available evidence, studies were not excluded based on their quality or level of evidence.

A meta-analysis was not conducted because too few studies reported quantitative results and confidence intervals.

## Results

### Study selection

The search yielded 713 results, 427 remained after duplicates were removed. Screening left 107 articles to be assessed for eligibility. After revision of full length articles, 18 remained for inclusion and data extraction. Figure 1 shows our process of study selection.

### Characteristics of included studies

Methodological characteristics of included studies are summarised in Table 1. When specified, the distribution of male and female participants was not significantly different. The average age of diagnosis was only reported in three studies and ranged from 3;6 to 4;4 years with overall ranges from 0;1 to 11;0 years (Oji *et al.*, 2013; Park *et al.*, 2000; ten Dam *et al.*, 2013). Four studies compared outcomes from the non-syndromic SMCP population to those with syndromic SMCP, most commonly velocardiofacial syndrome (VCFS) (Bezuhly *et al.*, 2012; Brandao *et al.*, 2011; Ng *et al.*, 2015; Seagle *et al.*, 2016). In two of these four studies, the non-syndromic population acted as controls to the syndromic group (Bezuhly *et al.*, 2012; Brandao *et al.*, 2011). Remaining studies compared outcomes based on surgery (Afrooz *et al.*, 2015; Ezzat *et al.*, 2016; Park *et al.*, 2000), cleft type (Park *et al.*, 2016; Schönweiler *et al.*, 1999; Seagle *et al.*, 2016) and age at surgery (Kwon *et al.*, 2018; Swanson *et al.*, 2017). Nine studies examined only non-syndromic SMCP groups (Baek *et al.*, 2017; Calis *et al.*, 2018; Calnan, 1954; Isotalo *et al.*, 2007; Kwon *et al.*, 2018; Oji *et al.*, 2013; Seagle *et al.*, 1999; Swanson *et al.*, 2017; ten Dam *et al.*, 2013). Eight studies documented anatomical features at time of diagnosis, however these were unspecific in two studies (Ng *et al.*, 2015; Oji *et al.*, 2013).

In addition to hypernasality reported in most studies, pre-diagnosis symptoms included articulation difficulties (ten Dam *et al.*, 2013), recurrent otitis media with effusion (Seagle *et al.*, 1999), conductive hearing loss (ten Dam *et al.*, 2013) and feeding problems (ten Dam *et al.*, 2013). Pre-diagnosis treatment included using a baby bottle mouth piece,

adenoidectomy, tonsillectomy, ventilation tubes and speech therapy (Seagle *et al.*, 1999; ten Dam *et al.*, 2013). Schönweiler *et al.* (1999) also reported pre-assessment orthodontic treatment for the broader cleft lip and/or palate group, however details specific to SMCP were not discussed.

Of the 18 studies included, one was a randomised controlled trial (level 1), nine were retrospective cohort studies (level 2), four were before-after treatment studies (level 3) and four were non-comparative case series' (level 3). The retrospective nature of most included studies allowed researchers to include larger numbers of participants, with six papers reporting data for 50 or more participants (Baek *et al.*, 2017; Bezuhly *et al.*, 2012; Isotalo *et al.*, 2007; Park *et al.*, 2000; Park *et al.*, 2016; Swanson *et al.*, 2017). Refer to Table 1 for graded levels of evidence. Level 1 or 2 studies that reported rater agreement and/or quantitative speech measures were assigned a "+" quality rating. Rater reliability or agreement was addressed in four studies (Baek *et al.*, 2017; Brandao *et al.*, 2011; Park *et al.*, 2000; Swanson *et al.*, 2017) and quantitative speech outcomes in six studies (Baek *et al.*, 2017; Bezuhly *et al.*, 2012; Brandao *et al.*, 2011; Calis *et al.*, 2018; Kwon *et al.*, 2018; Ng *et al.*, 2015). Studies with less than half of the checklist criteria fulfilled were assigned a "-" quality rating.

### **Outcome measurement tools**

Speech and language outcomes were measured using a range of methods relevant to the study country's spoken language. Table 2 shows a summary of included features. Hyponasality was also measured in two studies on a 10-degree scale (Seagle *et al.*, 1999; Seagle *et al.*, 2016). Where reported, perceptual speech assessment was completed by speech pathologists, with the exception of two studies where examiners were speech pathologists and cleft/craniofacial surgeons (Afrooz *et al.*, 2015), and plastic surgeons and medical students (Park *et al.*, 2016). Complementary nasoendoscopy and multiview videofluoroscopy results were reported in seven studies (Bezuhly *et al.*, 2012; Brandao *et al.*, 2011; Calis *et al.*, 2018; Calnan, 1954; Ezzat *et al.*, 2016; Ng *et al.*, 2015; Seagle *et al.*, 1999). See Table SII (online supporting information) for additional details of perceptual and instrumental measures (i.e., formal scales, speech samples, examiner agreement, and direct VPI measures).

## Speech features

Extracted outcome data shows speech characteristics only (Table 3), as no studies reported language outcomes specific to SMCP. To minimise the impact of confounding factors associated with surgery, pre-surgical characteristics are shown separately. Studies only reporting post-operative results were excluded from this table. Despite reporting pre-operative nasalance scores, data from Calis *et al.* (2018) were excluded here as results were not independently comparable to other studies – i.e., they were reported separately for each syllable and no summative or normative values were included. Phonology and language data reported in Schönweiler *et al.*'s (1999) study were also excluded as they were grouped with other forms of orofacial clefting and not specific to SMCP. In two studies, non-syndromic and syndromic cases were in the same experimental group, however individual participant data were reported, enabling relevant non-syndromic data to be extracted (Afrooz *et al.*, 2015; Oji *et al.*, 2013).

While speech characteristics varied across included studies, participants consistently presented with hypernasality and articulatory difficulties associated with VPI. Articulatory errors were broadly described as present or absent (ten Dam *et al.*, 2013) and non-oral or passive (Baek *et al.*, 2017; Calnan, 1954). Park *et al.* (2000) reported glottal and palatalised articulation errors in 40% and 13% of their study population respectively. One study found that 46 (62%) participants mis-articulated at least one of three alveolar sounds (/s, l, r/) independent of overall velopharyngeal functioning (Isotalo *et al.*, 2007). Despite not reporting pre-operative results, it is interesting to note that Schönweiler *et al.* (1999) found a positive linear correlation ( $p < 0.01$ ) between nasal resonance/NAE and posterior mis-articulation, suggesting that posterior articulatory patterns are a reflection of VPI.

Pre-surgical NAE was discussed in two papers (Ezzat *et al.*, 2016; Swanson *et al.*, 2017). Ezzat and colleagues reported that NAE was visible, audible or turbulent for 11 (55%) participants. Swanson *et al.* reported a mean NAE score of 2.0 and 1.5 for groups where participants received primary surgical repair at 3-3;9 years and 4+ years respectively, indicating that NAE was present and visible. Hyponasality was not reported in any studies. Three studies reported pre-operative nasalance scores, all showing abnormal hypernasal

resonance based on individually appropriate cutoff scores (Bezuhly *et al.*, 2012; Brandao *et al.*, 2011; Ng *et al.*, 2015).

All nasoendoscopy and multiview videofluoroscopy results were used to guide surgical planning and measure post-surgical outcomes. No studies demonstrated an association between direct instrumental measures and speech outcomes. Refer to Table SIII (online supporting information) for pre-operative nasoendoscopy and video fluoroscopy outcomes.

Of the 18 included studies, 13 only included participants who had undergone primary surgical repair, increasing the likelihood that they presented with the outcome – speech difficulties – at the time of assessment. Proportion data from these studies should be considered as descriptive rather than representative. In the remaining five studies, 6.1% to 39.4% of participants were asymptomatic and did not require surgery (Calnan, 1954; Isotalo *et al.*, 2007; Oji *et al.*, 2013; Swanson *et al.*, 2017; ten Dam *et al.*, 2013).

#### **Factors impacting speech outcomes**

Table 4 addresses the second aim and illustrates factors shown to impact outcomes. As there were no language outcomes, this section focuses on speech and resonance. Only post-operative outcomes are considered as no studies overtly commented on the impact of pre-surgical characteristics, such as hearing ability.

All studies measuring pre- and post-operative speech results showed that primary palatal repair improved overall velopharyngeal functioning, specifically resonance and NAE. The effect of surgical repair on articulation was inconclusive, with one study showing a significant decrease in glottal articulation (Park *et al.*, 2000) and another showing no significant improvement in overall articulation post-operatively (ten Dam *et al.*, 2013). Perceptually normal speech post-operatively was achieved for 12-88% of participants and an improvement was seen in 44-100% across all studies.

Evidence supporting the impact of surgery type on post-operative speech outcomes was mixed (Afrooz *et al.*, 2015; Calis *et al.*, 2018; Ezzat *et al.*, 2016; Park *et al.*, 2000). Afrooz *et al.* (2015) and Ezzat *et al.* (2016) reported no significant difference in post-

operative velopharyngeal functioning between 1) radical intravelar veloplasty and modified Furlow palatoplasty; and 2) radical intravelar veloplasty and V-Y pushback pharyngoplasty with intravelar veloplasty respectively. Park and colleagues (2000) found no significant difference in post-operative articulation between those who received a pushback palatoplasty, pharyngeal flap, pushback with pharyngeal flap or Furlow palatoplasty. However, they reported that participants receiving a pharyngeal flap achieved greater overall velopharyngeal functioning when compared to those who received a pushback palatoplasty. Calis *et al.* (2018) reported mixed nasalance results for Furlow palatoplasty and posterior pharyngeal flap with intravelar veloplasty across different speech stimuli, with an overall favour for the latter procedure.

A child's age at surgery had a significant impact on post-operative speech (Baek *et al.*, 2017; Bezuhly *et al.*, 2012; Kwon *et al.*, 2018; Oji *et al.*, 2013; Swanson *et al.*, 2017). A lower age at surgery consistently resulted in better speech outcomes. One study showed that overall velopharyngeal functioning improved significantly for a group of participants who received primary repair between three and four years but not for those after four years (Swanson *et al.*, 2017). Another study found that up to 65 months, with every month increase in age at surgery the odds of having residual post-operative hypernasality increased by a multiple of 1.02 (Baek *et al.*, 2017).

Pre-operative presentation and the surgeon completing a child's primary repair were also shown to impact post-operative speech outcomes (Baek *et al.*, 2017; Bezuhly *et al.*, 2012). Lower pre-operative nasalance scores and greater pre-operative nasopharyngeal closure ratings resulted in preferable perceptual and instrumental speech scores post-operatively (Bezuhly *et al.*, 2012). Pre-operative compensatory articulation was not shown to impact post-operative hypernasality, regardless of age at surgery (Baek *et al.*, 2017). The impact of primary surgeon should be considered with caution, as study authors suggested that outlying results were likely secondary to a lower number of surgical procedures completed by one of five surgeons (Bezuhly *et al.*, 2012).

One study showed that sex does not impact alveolar articulation post-operatively (Isotalo *et al.*, 2007). The impact of velocardiofacial syndrome on pre- and post-operative

speech outcomes differed across two studies (Bezuhly *et al.*, 2012; Brandao *et al.*, 2011). Brandao *et al.* (2011) reported no significant difference in perceptual or nasometric measures of velopharyngeal functioning. However, Bezuhly *et al.* (2012) found that a significantly greater proportion of individuals with non-syndromic SMCP achieved normal resonance – measured perceptually and instrumentally – when compared to those with velocardiofacial syndrome. They further reported a significant difference in the median time required to achieve normal resonance between the two groups.

## **Discussion**

This study aimed to systematically review and document the speech and language features of individuals with unrepaired non-syndromic SMCP. The study also sought to identify factors shown to impact speech or language outcomes amongst this population. Knowledge and understanding of all contributing factors is crucial to inform accurate diagnosis and guide treatment. We applied rigorous methodological processes to identify 18 eligible studies for inclusion. Included studies showed that individuals with non-syndromic SMCP may present with speech difficulties including hypernasality, nasal air emissions and disordered articulation. Speech outcomes varied with age at primary surgical repair, type of surgical procedure and the presence of an underlying syndrome. This review highlights a dearth of literature exploring motor speech, phonology and language abilities amongst individuals with non-syndromic SMCP.

### **Speech and language features**

Similar to the overt cleft palate population, this review suggests that individuals with unrepaired non-syndromic SMCP may present with speech difficulties secondary to VPI. The proportion of asymptomatic participants with SMCP across studies (6.1-39.4%) lies within the mid-range of previously reported figures (Crikelair *et al.*, 1970; Shprintzen *et al.*, 1985; Velasco *et al.*, 1988; Weatherley-White *et al.*, 1972). Most participants presented with mild to moderate hypernasality, a key diagnostic feature. Cleft related articulation errors were reported in consistently more than 40% of study participants (Baek *et al.*, 2017; Calnan, 1954; Park *et al.*, 2000). One study also reported alveolar articulatory errors that were

independent of overall velopharyngeal functioning, validating the results of a previous study (Isotalo *et al.*, 2007; Pulkkinen *et al.*, 2001). This observation may be of interest to the clinician, who can consider treating alveolar errors independent of VPI. However, as these results were only reported in one study, further exploration of alveolar errors in other non-syndromic SMCP populations is required before a definitive association can be made.

This review identifies the sparsity of literature outlining motor speech, phonological and language abilities of individuals with non-syndromic SMCP. Considering the emerging evidence suggesting language difficulties amongst the broader cleft lip and palate phenotype, investigations amongst the SMCP population appear timely and beneficial (Gallagher *et al.*, 2017; Hardin-Jones *et al.*, 2014; Klintö *et al.*, 2015; Knight *et al.*, 2015; Wehby *et al.*, 2014; Young *et al.*, 2012).

### **Factors impacting speech outcomes**

As surgical intervention was central to all studies, factors impacting post-operative speech results were considered in this review. Available evidence suggested that a lower age at surgery resulted in better post-operative speech outcomes. Evidence surrounding the impact of surgery type is inconclusive. Further investigations into the impact of the following factors is required before an informed judgement can be made: surgeon completing the primary repair; sex; presence of an underlying syndrome; and pre-operative articulation, nasalance and nasopharyngeal closure rating. Other potential pre- and post-surgical confounding factors not discussed amongst these studies include hearing ability, presenting anatomical features and socioeconomic status, all of which have been shown to influence broader speech and language development (Blamey *et al.*, 2001; Chen *et al.*, 1996; Hoff, 2003; Moeller, 2000).

### **Study methodological considerations**

The absence of randomised controlled trials and reliance on cohort or case study methodology is in line with previous systematic reviews and surveys in the area of cleft lip and palate (de Ladeira *et al.*, 2012; Gilleard *et al.*, 2014; Karri, 2006). The retrospective nature of most studies is likely secondary to the complex nature of diagnosis and treatment

decision making within this population. As the primary purpose of this review was to identify descriptive speech and language features, including level 2 and 3 studies allowed valuable characteristic data to be considered. Results should also be considered in light of limitations associated with the non-specific SMCP and non-syndromic diagnostic criteria of some studies. Further, when determining eligibility, a substantial number of studies were excluded for combining non-syndromic and syndromic or cleft lip and/or palate and SMCP group outcomes, not allowing for the extraction of data specific to non-syndromic SMCP (e.g., Ettinger *et al.*, 2018; Mehendale *et al.*, 2003; Pet *et al.*, 2015; Rise, 1966; Roberts *et al.*, 1983; Rogers *et al.*, 2013; Sie *et al.*, 2001; Sommerlad *et al.*, 2004; Ysunza *et al.*, 2011). While this distinction was important to determine the isolated impact of SMCP, it limited the number of studies that could be included. Precise descriptions of SMCP and non-syndromic diagnoses are becoming increasingly significant as contemporary definitions and grading systems continue to evolve (Dixon *et al.*, 2011; Smyth, 2014; Sommerlad *et al.*, 2004).

### Conclusion

Fifteen of the 18 studies included in this review are retrospective reviews, representing varying levels of evidence and risk of bias. However, similar patterns of speech outcomes were found across all studies. Individuals with unrepaired non-syndromic SMCP may present with mild to severe hypernasality, cleft speech articulation errors and nasal air emissions. This review highlights a paucity of research examining motor or phonological aspects of speech and receptive or expressive language skills amongst this population. A lower age at surgery was associated with better post-operative velopharyngeal functioning. Evidence supporting other impacting factors is limited and reliable conclusions cannot be made.

Clinically, this study clarifies that a relatively high proportion of individuals with unrepaired non-syndromic SMCP have speech difficulties associated with VPI. Speech features are similar to those of individuals with cleft palate and form a key pre-diagnosis symptom. Characteristics such as non-oral or weak articulation, nasal air emissions or hypernasality may be indicative of palatal dysfunction. The speech language therapist holds a key role in identifying these difficulties and pursuing further investigations for SMCP

diagnoses where appropriate. Other pre-diagnosis symptoms include recurrent otitis media, hearing loss and feeding problems. Therefore, the wider care team – including primary care physicians, nurses and specialists – also has a role in recognising symptoms and conducting visual inspections of the palate to facilitate early diagnosis. In addition to velopharyngeal insufficiency, speech and language therapists may look for alveolar articulation errors and consider treating these independently. Where possible, earlier surgical intervention is preferred for better speech outcomes. Further evidence is required to validate a definitive age at which primary surgical repair improves or worsens outcomes.

Further research, examining broader aspects of speech and language skills in this population is required. We recommend that inter- and intra-examiner reliability is always completed and reported, along with quantitative instrumental speech assessment (e.g., nasometry). Researchers should consider and measure confounding factors that are likely to impact pre- and post-surgical outcomes. With all the above investigations, age at diagnosis, pre-diagnostic symptoms and pre-surgical anatomical features should be clearly defined. Evidence exploring all areas of communication and their impacting factors can facilitate earlier clinical screening and informed holistic clinical treatment planning.

### **Key Messages**

- Individuals with unrepaired SMCP may have articulatory and resonatory difficulties associated with velopharyngeal insufficiency.
- A lower age at primary palatal surgical repair appears to lead to better post-surgical speech outcomes.
- This review identifies a lack of research exploring language characteristics and phonological speech patterns of individuals with non-syndromic SMCP.



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**Figure 1. PRISMA process of study selection**

Table 1. Methodological characteristics of included studies

Study (first author)	Demographic			SMCP		SIGN <sup>a</sup>	
	N	Mean age at diagnosis (y)	Comparison group	Key anatomical features (overt or occult)	Exclusion criteria	Study design	Level of evidence
Afroz, 2015 <sup>b</sup>	25	NR	Different surgery type	Occult: Lack of classic Calnan triad; inverted V-shape in soft palate elevation; anterior displacement of uvula during phonation	Complete or incomplete CP; overt SMCP; speech abnormalities secondary to idiopathic palatal paresis; global hypotonia; severe developmental delay	Cohort study (retrospective)	2-
Baek, 2017	74	NR	NA	Overt: Range one (n=13), two (n=31) or all (n=24) of Calnan's triad. Occult: Lack of classic Calnan triad (n=6)	Syndromic; underlying disorders; congenital hearing loss; hemifacial microsomia	Before-after study (retrospective)	3
Bezuhly, 2012 <sup>b</sup>	55	NR	VCFS	Overt: All features of Calnan triad Occult: Without all three features of Calnan triad, hypoplastic or absent uvular ridge, flattened velum, malfunction on nasopharyngoscopy	Hypodynamic velum without discernible anatomical abnormality	Cohort study (retrospective)	2+
Brandao, 2011 <sup>b</sup>	25	NR	VCFS	NR	NR Inclusion criteria listed, but not exclusion	Cohort study (retrospective)	2+
Calis, 2018	29	NR	NA	NR	Developmental delay; mental retardation; hearing loss; syndromic appearance; without follow up or pre-operative records; <4 years; secondary surgery.	Before-after study (retrospective)	3
Calnan, 1954 <sup>b</sup>	13	NR	NA	NR <sup>c</sup>	NR	Non-comparative case series	3
Ezzat, 2016	20	NR	Different surgery type	NR	Syndromic; responding to speech therapy; undergone previous surgery for SMCP; intellectual disability	Individual randomised trial	1-

Isotalo, 2007	58	NR	NA	NR	NR	Inclusion criteria listed, but not exclusion	Non-comparative case series (retrospective)	3
Kwon, 2018	23 <sup>d</sup>	NR	NA	NR	Syndromic		Non-comparative case series (retrospective)	3
Ng, 2015 <sup>b</sup>	17	NR	Syndromic	Overt: Calnan triad (details not specified)	NR	Inclusion criteria listed, but not exclusion	Cohort study (retrospective)	2+
Oji, 2013 <sup>b</sup>	9	5;8	NA	Bifid uvula (details not specified)	NR		Non-comparative case series (retrospective)	3
Park, 2000	50	3;9	Different surgery type	Overt: Obvious midline muscular diastasis and bifid uvula	Adequate speech; neurological deficits; severe developmental delay; hearing loss; short or no follow up		Cohort study (retrospective)	2+
Park, 2016	53	NR	ICP	NR	Syndromic; > 10 years		Cohort study (retrospective)	2-
Schönweiler, 1999	12	NR	Cleft type	NR	CLO; syndromic; developmental delay; bilateral sensorineural hearing loss		Cohort study (retrospective)	2-
Seagle, 1999	29	NR	NA	Overt and occult: Range, none to all of Calnan's triad	Neurological deficits; significant hearing loss; syndromic diagnosis precluding normal speech development		Before-after study (retrospective)	3
Seagle, 2016 <sup>b</sup>	15	NR	NS cleft types; syndromic; non-cleft VPI	NR	Syndromic		Cohort study (retrospective)	2-
Swanson, 2017	66	NR	Age at surgery	NR	Syndromic; without complete medical record		Cohort study (retrospective)	2+
ten Dam,	28	3;7(1;9)	NA	Overt and Occult: Range, none to all: Calnan	Syndromic; additional comorbidity; lack of		Before-after study	3

2013

triad

follow-up

(retrospective)

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<sup>a</sup> Determined using the SIGN *Algorithm for classifying study design for questions of effectiveness* <http://www.sign.ac.uk/methodology/>. <sup>b</sup> These studies included data from both syndromic and non-syndromic cases, only data on non-syndromic participants are included here. <sup>c</sup> Anatomical features were reported for some individual cases (including bifid uvula and palatal notch), however a group summary was not included. <sup>d</sup> Speech data were only available for 20 participants.

NR = not reported. NA = not applicable. NS = non-syndromic. CP = cleft palate. ICP = incomplete cleft palate. CLO = cleft lip only. SMCP = submucous cleft palate. VCFS = velocardiofacial syndrome. VPI = velopharyngeal insufficiency. SIGN = Scottish Intercollegiate Guidelines Network.

Table 2. Speech and language outcome measures

Study (first author)	Speech measures						
	Perceptual assessment				Raters (N)	Instrumental assessment <sup>a</sup>	Language measure
	Hypernasality scale	NAE scale	Articulation scale	Overall VP function			
Afroz, 2015	NR	NR	NR	Score out of 35	NR	NA	NA
Baek, 2017	4 degrees	2 degrees	Descriptive <sup>b</sup>	4 degrees	1 of 2	Nasometry – oral passages and nasal sentences	NA
Bezuhly, 2012	4 degrees	NR	NR	NR	1	Nasometry – bilabial plosive syllable repetition /pa pa pa/	NA
Brandao, 2011	6 degrees <sup>c</sup>	NR	NR	NR	3	Nasometry – Brazilian Portuguese oral sentences	NA
Calis, 2018	NA	NA	NA	NA	NA	Nasometry – syllables	NA
Calnan, 1954	5 degrees	4 degrees	NR	NR	NR	NA	NA
Ezzat, 2016	NR	2 degrees	4 degrees	NR	1	NA	NA
Isotalo, 2007	4 degrees	NR	Descriptive <sup>b</sup>	4 degrees	1	Nasometry – standardised Finnish sentences (results not reported)	NA
Kwon, 2018	NR	NR	NR	NR	NA	Nasometry – vowels, words and sentences (details not provided)	NA
Ng, 2015	4 degrees	NR	NR	NR	1 of 3	Nasometry – passage not specified	NA
Oji, 2013	NR	NR	NR	4 degrees	2	NR	NA

Park, 2000	4 degrees	4 degrees	Descriptive <sup>b</sup>	7 degrees	3	NA	NA
Park, 2016	NR	NR	NR	Score out of 66	10	NA	NA
Schönweiler, 1999	4 degrees	4 degrees	NR	NA	NR	NA	Sentence structure; vocabulary; receptive language; auditory discrimination; auditory memory <sup>d</sup>
Seagle, 1999	10 degrees	10 degrees	Descriptive <sup>c</sup>	2 degrees	1 of 2	NA	NA
Seagle, 2016	10 degrees	10 degrees	NR	2 degrees	NR	NA	NA
Swanson, 2017	5 degrees	3 degrees	5 degrees	8 degrees	1 of 2	NA	NA
ten Dam, 2013	3 degrees	NR	2 degrees	NR	1	NA	NA

<sup>a</sup> Details of nasoendoscopic and multiview videofluoroscopic measures are outlined in the text and supplementary tables. <sup>b</sup> Presence or absence of particular articulatory patterns (e.g., mis-articulation of alveolar articulation (/r, s, l/), substitution of glottal stops, palatalisation, omission, nasal air emission, compensatory articulation). <sup>c</sup> Scale reduced to 4 degrees for analysis. <sup>d</sup> Results not specific to SMCP and are not discussed further.

NR = not reported. NA = not applicable. VP = velopharyngeal. NAE = nasal air emissions.

Table 3. Pre-operative speech outcomes

Study			Perceptual speech measures			Instrumental measure
(first author)	Hearing status	Speech therapy (outcome) <sup>a</sup>	Hypernasality	Articulation	Overall VP function	Mean nasalance
Afrooz, 2015	NR	Yes (not effective)	NR	NR	25 incompetent VP functioning (100%, PWSS >7)	NA
Baek, 2017	No congenital hearing loss	Yes <sup>b</sup>	NR	33 compensatory articulation (45%) 32 no compensatory articulation (43%) 9 no data (12%)	NR	NR
Bezuhly, 2012	9 hearing loss (16%)	Yes (not effective)	37 mild (67%) 15 moderate (27%) 3 severe (5%)	NR	NR	Abnormal (hypernasal)
Brandao, 2011	NR	NR	6 mild (24%) 14 moderate (56%) 5 severe (20%)	NR	NR	Abnormal (hypernasal)
Calnan, 1954	NR	NR	NR	9 Group I (69%) 3 Group II (23%) 1 Group III (8%) <sup>c</sup>	NR	NA
Ezzat, 2016	NR	Yes (not effective)	12 mild (60%) 5 moderate (25%) 3 severe (15%)	NR	NR	NA
Isotalo, 2007	No persistent hearing loss	Yes (NR)	See overall	46 at least one sound correctly articulated (62%) 2 all sounds misarticulated: (3%) <sup>d</sup>	2 typical (3%) 1 audible NAE (2%) 19 hypernasality +/- audible NAE (33%) 36 weak plosives or compensatory articulation +/- hypernasality and NAE (62%)	NR

Ng, 2015	7 hearing loss (41%) <sup>c</sup>	Yes (not effective)	9 mild (53%) 7 moderate (41%) 1 severe (6%)	NR	NR	Abnormal (hypernasal)
Oji, 2013	NR	NR	NA	NA	1 typical (11%) 7 moderate (78%) 1 severe (11%)	NA
Park, 2000 <sup>f</sup>	WNL	Yes (not effective)	NR	18 glottal articulation (40%) 6 palatalised articulation (13%)	0 typical 10 fair (22%) 35 poor (78%)	NA
Swanson, 2017	2 mild conductive hearing loss (6%) <sup>g</sup>	NR	Unrepaired: 0.17(0.4) normal Repair 3-3;9: 2.5(1.7) moderate Repair 4+: 2.8(0.96) moderate <sup>h</sup>	Unrepaired: 1 (1.5) Repair 3-3;9: 4.8 (3.3) Repair 4+: 1.8 (3.5) <sup>hi</sup>	Unrepaired: 3.0(1.8), borderline Repair 3-3;9: 9.5(5.8), incompetent Repair 4+: 6.5(3.7) incompetent <sup>h</sup>	NA
ten Dam, 2013	11 conductive hearing loss (45.8%)	Yes (NR)	5 normal (22%) 18 hypernasal (78%)	10 normal articulation (44%) 13 articulation problems (56%)	NR	NA

<sup>a</sup> Pre-operative. <sup>b</sup> Speech therapy offered pre-operatively if there was a pinhole-sized velopharyngeal gap and intermittent hypernasality. <sup>c</sup> Group I=Passive errors; Group II=Non-oral errors; Group III=Unintelligible, all consonant sounds substituted for glottal stops or vowel like sounds (Wardill, 1933). <sup>d</sup> Alveolar sounds /s, l, r/. <sup>e</sup> Reported to not affect speech outcomes. <sup>f</sup> Pre-operative speech outcomes were reported for 45 patients. <sup>g</sup> Hearing data only available for 33 participants. <sup>h</sup> Pre-operative speech results were reported as component PWSS scores by timing of repair: repair 3-3;9 years; repair 4+ years; unrepaired. Overall data were not reported. Figures reported here are means within each group. Note that pre-surgical data for those repaired <3 years were not measured. <sup>i</sup> Summative Pittsburgh Weighted Speech Score out of 23.

NR = not reported. NA = not applicable. WNL = within normal limits. VP = velopharyngeal. PWSS = Pittsburgh Weighted Speech Scale.

Table 4. Factors impacting post-surgical speech outcomes

Surgery type	§β	++	Pre-operative compensatory articulation	Ω	-
	§βΩ†	----			
Primary surgeon	Ω	+	Pre-operative nasalance score	Ω§	+
Age at surgery	§Ωβ	++++	Pre-operative nasopharyngeal closure rating	Ω§	+
Sex	†	-	Syndrome	Ω§	+/-

Ω perceptual ratings of resonance and/or nasal air emissions

§ nasalance scores

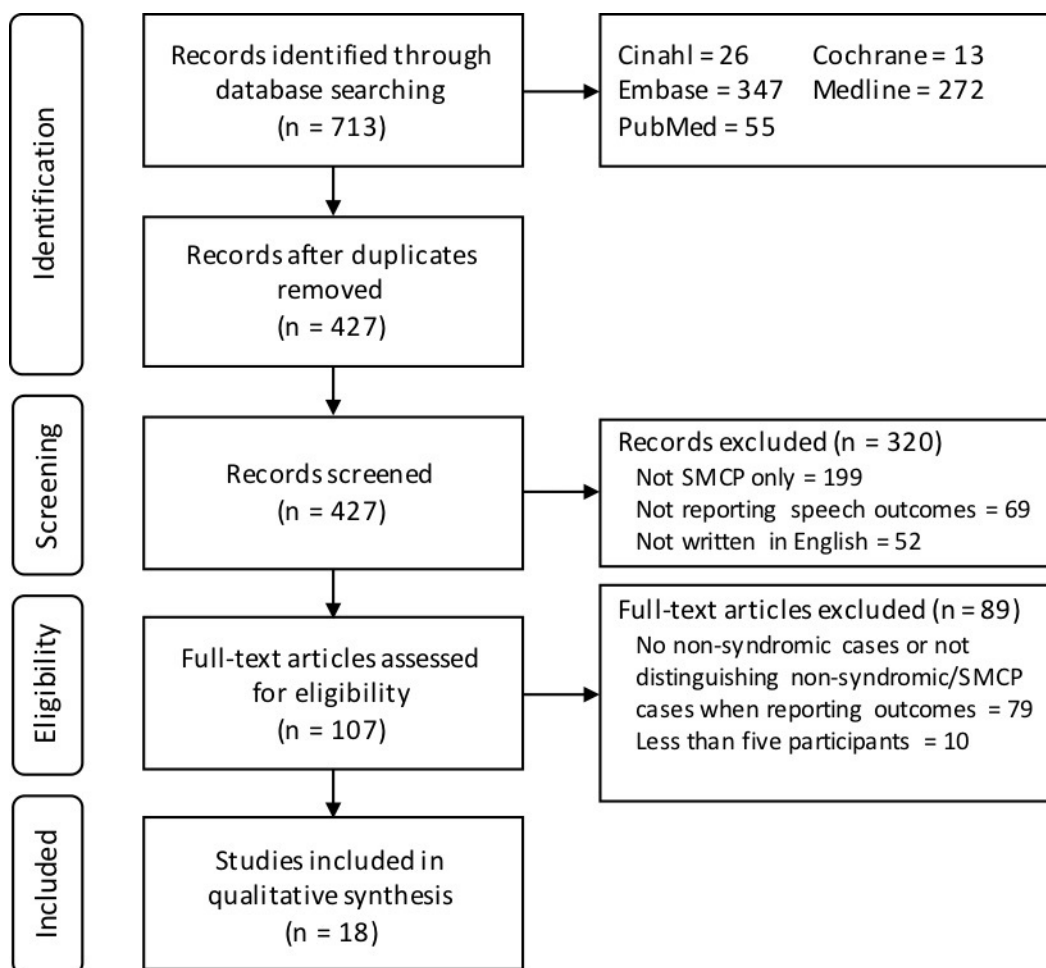
† perceptual ratings of articulation

β cumulative speech score

∂ phonology

+ /++ /+++ /++++ significant impact reported by 1/2/3/4+ studies with  $p < 0.05$

- /-- no significant impact reported by 1/2 studies with  $p > 0.1$



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