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.
References


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Figure 1. PRISMA process of study selection
<table>
<thead>
<tr>
<th>Study (first author)</th>
<th>N</th>
<th>Mean age at diagnosis (y)</th>
<th>Comparison group</th>
<th>Demographic</th>
<th>SMCP</th>
<th>Key anatomical features (overt or occult)</th>
<th>Exclusion criteria</th>
<th>Study design</th>
<th>Level of evidence</th>
</tr>
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<tbody>
<tr>
<td>Afrooz, 2015b</td>
<td>25</td>
<td>NR</td>
<td>Different surgery type</td>
<td></td>
<td></td>
<td>Occult: Lack of classic Calnan triad; inverted V-shape in soft palate elevation; anterior displacement of uvula during phonation</td>
<td>Complete or incomplete CP; overt SMCP; speech abnormalities secondary to idiopathic palatal paresis; global hypotonia; severe developmental delay</td>
<td>Cohort study (retrospective)</td>
<td>2-</td>
</tr>
<tr>
<td>Back, 2017</td>
<td>74</td>
<td>NR</td>
<td>NA</td>
<td></td>
<td></td>
<td>Overt: Range one (n=13), two (n=31) or all (n=24) of Calnan’s triad. Occult: Lack of classic Calnan triad (n=6)</td>
<td>Syndromic; underlying disorders; congenital hearing loss; hemifacial microsomia</td>
<td>Before-after study (retrospective)</td>
<td>3</td>
</tr>
<tr>
<td>Bezuhly, 2012b</td>
<td>55</td>
<td>NR</td>
<td>VCFS</td>
<td></td>
<td></td>
<td>Overt: All features of Calnan triad Occult: Without all three features of Calnan triad, hypoplastic or absent uvular ridge, flattened velum, malfunction on nasopharyngoscopy</td>
<td>Hypodynamic velum without discernible anatomical abnormality</td>
<td>Cohort study (retrospective)</td>
<td>2+</td>
</tr>
<tr>
<td>Brandao, 2011b</td>
<td>25</td>
<td>NR</td>
<td>VCFS</td>
<td></td>
<td></td>
<td>NR</td>
<td>NR</td>
<td>Cohort study (retrospective)</td>
<td>2+</td>
</tr>
<tr>
<td>Calis, 2018</td>
<td>29</td>
<td>NR</td>
<td>NA</td>
<td></td>
<td></td>
<td>NR</td>
<td>Developmental delay; mental retardation; hearing loss; syndromic appearance; without follow up or pre-operative records; ≤4 years; secondary surgery.</td>
<td>Before-after study (retrospective)</td>
<td>3</td>
</tr>
<tr>
<td>Calnan, 1954b</td>
<td>13</td>
<td>NR</td>
<td>NA</td>
<td></td>
<td></td>
<td>NR</td>
<td>NR</td>
<td>Non-comparative case series</td>
<td>3</td>
</tr>
<tr>
<td>Ezzat, 2016</td>
<td>20</td>
<td>NR</td>
<td>Different surgery type</td>
<td></td>
<td></td>
<td>NR</td>
<td>Syndromic; responding to speech therapy; undergone previous surgery for SMCP; intellectual disability</td>
<td>Individual randomised trial</td>
<td>1-</td>
</tr>
<tr>
<td>Reference</td>
<td>Age</td>
<td>Sex</td>
<td>Diagnosis</td>
<td>Clinical Presentation</td>
<td>Study Design</td>
<td>Quality Score</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>----------------------------</td>
<td>-----</td>
<td>-----</td>
<td>--------------------------------</td>
<td>---------------------------------------------------------------------------------------</td>
<td>----------------------------------</td>
<td>---------------</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Isotalo, 2007</td>
<td>NR</td>
<td>NA</td>
<td>NR</td>
<td>Inclusion criteria listed, but not exclusion</td>
<td>Non-comparative case series</td>
<td>3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Kwon, 2018</td>
<td>23</td>
<td>NR</td>
<td>NR</td>
<td>Syndromic</td>
<td>Non-comparative case series</td>
<td>3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Ng, 2015&lt;sup&gt;b&lt;/sup&gt;</td>
<td>17</td>
<td>NR</td>
<td>Syndromic</td>
<td>Inclusion criteria listed, but not exclusion</td>
<td>Cohort study (retrospective)</td>
<td>2+</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Oji, 2013&lt;sup&gt;b&lt;/sup&gt;</td>
<td>9</td>
<td>5;8</td>
<td>NA</td>
<td>Bifid uvula (details not specified)</td>
<td>Non-comparative case series</td>
<td>3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Park, 2000</td>
<td>50</td>
<td>3:9</td>
<td>Different surgery type</td>
<td>Adequate speech; neurological deficits; severe developmental delay; hearing loss;</td>
<td>Cohort study (retrospective)</td>
<td>2+</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Park, 2016</td>
<td>53</td>
<td>NR</td>
<td>ICP</td>
<td>Syndromic; &gt; 10 years</td>
<td>Cohort study (retrospective)</td>
<td>2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Schönweiler, 1999</td>
<td>12</td>
<td>NR</td>
<td>Cleft type</td>
<td>CLO; syndromic; developmental delay; bilateral sensorineural hearing loss</td>
<td>Cohort study (retrospective)</td>
<td>2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Seagle, 1999</td>
<td>29</td>
<td>NR</td>
<td>NA</td>
<td>Neurological deficits; significant hearing loss; syndromic diagnosis precluding normal speech development</td>
<td>Before-after study (retrospective)</td>
<td>3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Seagle, 2016&lt;sup&gt;b&lt;/sup&gt;</td>
<td>15</td>
<td>NR</td>
<td>NS cleft types; syndromic; non-cleft VPI</td>
<td>Syndromic</td>
<td>Cohort study (retrospective)</td>
<td>2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Swanson, 2017</td>
<td>66</td>
<td>NR</td>
<td>Age at surgery</td>
<td>Syndromic; without complete medical record</td>
<td>Cohort study (retrospective)</td>
<td>2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ten Dam, 2017</td>
<td>28</td>
<td>3:7(1:9)</td>
<td>NA</td>
<td>Syndromic; additional comorbidity; lack of</td>
<td>Before-after study</td>
<td>3</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

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<table>
<thead>
<tr>
<th>2013</th>
<th>triad</th>
<th>follow-up</th>
<th>(retrospective)</th>
</tr>
</thead>
</table>

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

<table>
<thead>
<tr>
<th>Study (first author)</th>
<th>Hypernasality scale</th>
<th>NAE scale</th>
<th>Articulation scale</th>
<th>Overall VP function</th>
<th>Raters (N)</th>
<th>Instrumental assessment</th>
<th>Language measure</th>
</tr>
</thead>
<tbody>
<tr>
<td>Afrooz, 2015</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Score out of 35</td>
<td>NR</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Baek, 2017</td>
<td>4 degrees</td>
<td>2 degrees</td>
<td>Descriptive</td>
<td>4 degrees</td>
<td>1 of 2</td>
<td>Nasometry – oral passages and nasal sentences</td>
<td>NA</td>
</tr>
<tr>
<td>Bezuhly, 2012</td>
<td>4 degrees</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>1</td>
<td>Nasometry – bilabial plosive syllable repetition /pa pa pa/</td>
<td>NA</td>
</tr>
<tr>
<td>Brandao, 2011</td>
<td>6 degrees</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>3</td>
<td>Nasometry – Brazilian Portuguese oral sentences</td>
<td>NA</td>
</tr>
<tr>
<td>Calis, 2018</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>NA</td>
<td>Nasometry – syllables</td>
<td>NA</td>
</tr>
<tr>
<td>Calnan, 1954</td>
<td>5 degrees</td>
<td>4 degrees</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Ezzat, 2016</td>
<td>NR</td>
<td>2 degrees</td>
<td>4 degrees</td>
<td>NR</td>
<td>1</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Isotalo, 2007</td>
<td>4 degrees</td>
<td>NR</td>
<td>Descriptive</td>
<td>4 degrees</td>
<td>1</td>
<td>Nasometry – standardised Finnish sentences (results not reported)</td>
<td>NA</td>
</tr>
<tr>
<td>Kwon, 2018</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NA</td>
<td>Nasometry – vowels, words and sentences (details not provided)</td>
<td>NA</td>
</tr>
<tr>
<td>Ng, 2015</td>
<td>4 degrees</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>1 of 3</td>
<td>Nasometry – passage not specified</td>
<td>NA</td>
</tr>
<tr>
<td>Oji, 2013</td>
<td>NR</td>
<td>NR</td>
<td>4 degrees</td>
<td>2</td>
<td>NR</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Study</td>
<td>Degree</td>
<td>Degree</td>
<td>Type</td>
<td>Degree</td>
<td>Score</td>
<td>Out of 66</td>
<td>VP</td>
</tr>
<tr>
<td>-------------------</td>
<td>--------</td>
<td>--------</td>
<td>---------------</td>
<td>--------</td>
<td>-------</td>
<td>-----------</td>
<td>----</td>
</tr>
<tr>
<td>Park, 2000</td>
<td>4</td>
<td>4</td>
<td>Descriptive(^b)</td>
<td>7</td>
<td>3</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Park, 2016</td>
<td>NR</td>
<td>NR</td>
<td>Score out of 66</td>
<td>10</td>
<td>10</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Schönweiler, 1999</td>
<td>4</td>
<td>4</td>
<td>NR</td>
<td>NA</td>
<td>NR</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Seagle, 1999</td>
<td>10</td>
<td>10</td>
<td>Descriptive(^c)</td>
<td>2</td>
<td>1 of 2</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Seagle, 2016</td>
<td>10</td>
<td>10</td>
<td>NR</td>
<td>2</td>
<td>NR</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>Swanson, 2017</td>
<td>5</td>
<td>3</td>
<td>5</td>
<td>8</td>
<td>1 of 2</td>
<td>NA</td>
<td>NA</td>
</tr>
<tr>
<td>ten Dam, 2013</td>
<td>3</td>
<td>2</td>
<td>NR</td>
<td>1</td>
<td>1</td>
<td>NA</td>
<td>NA</td>
</tr>
</tbody>
</table>

\(^a\) NR = not reported. NA = not applicable. VP = velopharyngeal. NAE = nasal air emissions.
<table>
<thead>
<tr>
<th>Study (first author)</th>
<th>Hearing status</th>
<th>Speech therapy (outcome)</th>
<th>Perceptual speech measures</th>
<th>Instrumental measure</th>
</tr>
</thead>
<tbody>
<tr>
<td>Afrooz, 2015</td>
<td>NR</td>
<td>Yes (not effective)</td>
<td>NR</td>
<td>25 incompetent VP functioning (100%, PWSS &gt;7)</td>
</tr>
<tr>
<td>Baek, 2017</td>
<td>No congenital hearing loss</td>
<td>Yes</td>
<td>NR</td>
<td>33 compensatory articulation (45%)</td>
</tr>
<tr>
<td>Bezuhly, 2012</td>
<td>9 hearing loss (16%)</td>
<td>Yes (not effective)</td>
<td>NR</td>
<td>32 no compensatory articulation (43%)</td>
</tr>
<tr>
<td>Brandao, 2011</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>9 no data (12%)</td>
</tr>
<tr>
<td>Calnan, 1954</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>Abnormal (hypernasal)</td>
</tr>
<tr>
<td>Ezzat, 2016</td>
<td>NR</td>
<td>Yes (not effective)</td>
<td>NR</td>
<td>Abnormal (hypernasal)</td>
</tr>
<tr>
<td>Isotalo, 2007</td>
<td>No persistent hearing loss</td>
<td>Yes (NR)</td>
<td>See overall</td>
<td>46 at least one sound correctly articulated (62%)</td>
</tr>
</tbody>
</table>

<sup>a</sup> This article is protected by copyright. All rights reserved.
<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Type of Hearing Loss</th>
<th>Speech Outcomes</th>
<th>Articulation Problems</th>
<th>Abnormality</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ng, 2015</td>
<td></td>
<td>7 hearing loss (41%)</td>
<td>Yes (not effective)</td>
<td>9 mild (53%)</td>
<td>NR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>7 moderate (41%)</td>
<td></td>
<td>1 severe (6%)</td>
<td>NR</td>
</tr>
<tr>
<td>Oji, 2013</td>
<td></td>
<td>NR</td>
<td>NR</td>
<td>NA</td>
<td>NR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>NA</td>
<td>NA</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>Park, 2000</td>
<td></td>
<td>NR</td>
<td>Yes (not effective)</td>
<td>NR</td>
<td>18 glottal articulation (40%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>NR</td>
<td>6 palatalised articulation (13%)</td>
<td>0 typical</td>
<td>10 fair (22%)</td>
</tr>
<tr>
<td>Swanson, 2017</td>
<td></td>
<td>2 mild conductive hearing loss (6%)</td>
<td>NR</td>
<td>Unrepaired: 0.17 (0.4) normal</td>
<td>5 normal (22%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Repair 3-3; 9: 2.5 (1.7) moderate</td>
<td>Repair 3-3: 4.8 (3.3)</td>
<td>Repair 4+: 1.8 (3.5)</td>
<td>18 hypernasal (78%)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
<td>NR</td>
</tr>
<tr>
<td>ten Dam, 2013</td>
<td></td>
<td>11 conductive hearing loss (45.8%)</td>
<td>Yes (NR)</td>
<td>5 normal (22%)</td>
<td>NR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>18 hypernasal (78%)</td>
<td>10 normal articulation (44%)</td>
<td>NR</td>
<td>NR</td>
</tr>
</tbody>
</table>

- Pre-operative.
- Speech therapy offered pre-operatively if there was a pinhole-sized velopharyngeal gap and intermittent hypernasality.
- 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).
- Alveolar sounds /s, l, r/.
- Reported to not affect speech outcomes.
- Pre-operative speech outcomes were reported for 45 patients.
- Hearing data only available for 33 participants.
- 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.
- 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

<table>
<thead>
<tr>
<th>Surgery type</th>
<th>§β</th>
<th>++</th>
<th>Pre-operative compensatory articulation Ω</th>
<th>-</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>βΩ†</td>
<td>----</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Primary surgeon</td>
<td>Ω</td>
<td>+</td>
<td>Pre-operative nasalance score Ω§</td>
<td>+</td>
</tr>
<tr>
<td>Age at surgery</td>
<td>§Ωβ</td>
<td>+++</td>
<td>Pre-operative nasopharyngeal closure rating Ω§</td>
<td>+</td>
</tr>
<tr>
<td>Sex†</td>
<td>-</td>
<td>Syndrome Ω§</td>
<td>+/-</td>
<td></td>
</tr>
</tbody>
</table>

Ω perceptual ratings of resonance and/or nasal air emissions
§ nasalance scores
† perceptual ratings of articulation
β cumulative speech score
β̃ 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
Author/s:
Boyce, JO; Kilpatrick, N; Morgan, AT

Title:
Speech and language characteristics in individuals with nonsyndromic submucous cleft palate-A systematic review

Date:
2018-11-01

Citation:

Persistent Link:
http://hdl.handle.net/11343/284398