Scandcleft Randomized Trials of Primary Surgery for Unilateral Cleft Lip and Palate

Speech Proficiency at 10 Years of Age

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RESEARCH REPORT

Scandcleft randomized trials of primary surgery for unilateral cleft lip and palate: Speech proficiency at 10 years of age

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Abstract

Background & Aim: To assess consonant proficiency and velopharyngeal function in 10-year-old children born with unilateral cleft lip and palate (UCLP) within the Scandcleft project.

Methods & Procedures: Three parallel group, randomized, clinical trials were undertaken as an international multicentre study by nine cleft teams in five countries. Three different surgical protocols for primary palate repair (Arm B–Lip and soft palate closure at 3–4 months, hard palate closure at 36 months, Arm C–Lip closure at 3–4 months, hard and soft palate closure at 12 months, and Arm D–Lip closure at 3–4 months combined with a single-layer closure of the hard palate using a vomer flap, soft palate closure at 12 months) were tested against a common procedure (Arm A–Lip and soft palate closure at 3–4 months followed by hard palate closure at 12 months) in the total cohort of 431 children born with a non-syndromic UCLP. Speech audio and video recordings of 399 children were available and perceptually analysed. Percentage of consonants correct (PCC) from a naming test, an overall rating of velopharyngeal competence (VPC) (VPC-Rate), and a composite measure (VPC-Sum) were reported.

Outcomes & Results: The mean levels of consonant proficiency (PCC score) in the trial arms were 86–92% and between 58% and 83% of the children had VPC (VPC-Sum). Only 50–73% of the participants had a consonant proficiency level with their peers. Girls performed better throughout. Long delay of the hard palate repair (Arm B) indicated lower PCC and simultaneous hard and soft palate closure higher (Arm C). However, the proportion of participants with primary VPC (not including velopharyngeal surgeries) was highest in Arm B (68%) and lowest in Arm C (47%).

Conclusions & Implications: The speech outcome in terms of PCC and VPC was low across the trials. The different protocols had their pros and cons and there is no obvious evidence to recommend any of the protocols as superior. Aspects other than primary surgical method, such as time after velopharyngeal surgery, surgical experience, hearing level, language difficulties and speech therapy, need to be thoroughly reviewed for a better understanding of what has affected speech outcome at 10 years.

KEYWORDS

consonant proficiency, palatal surgical protocols, randomized controlled trial (RCT), unilateral cleft lip and palate (UCLP), velopharyngeal competence (VPC) velopharyngeal incompetence (VPI)

WHAT THIS PAPER ADDS

What is already known on the subject

- Speech outcomes at 10 years of age in children treated for UCLP are sparse and contradictory. Previous studies have examined speech outcomes and the relationship with surgical intervention in 5-year-olds.

What this study adds to the existing knowledge

- Speech outcomes based on standardized assessment in a large group of 10-year-old children born with UCLP and surgically treated according to different protocols are presented. While speech therapy had been provided, a large proportion of the children across treatment protocols still needed further speech therapy.

What are the potential or actual clinical implications of this work?

- Aspects other than surgery and speech function might add to the understanding of what affects speech outcome. Effective speech therapy should be available for children in addition to primary surgical repair of the cleft and secondary surgeries if needed.

INTRODUCTION

The evidence base to guide clinical teams in the selection of the best possible surgical procedure and surgical timing, taking account of both maxillary growth and speech development (Peterson-Falzone, 2013), is still weak (e.g., Reddy et al., 2017; Semb et al., 2017; Shaw et al., 2015). This lack of evidence was the motivation behind the international randomized clinical project Scandcleft. The primary outcomes of speech and dentofacial development after different surgical protocols in children born with unilateral cleft lip and palate (UCLP) were first investigated at 5 years of age (Semb et al., 2017). The focus of the present article is speech outcome at 10 years of age.

The Scandcleft project commenced in 1997. It compared the outcomes of different surgical protocols and included ten established cleft centres in five countries (Semb et al., 2017). Three randomized trials were started (Trials 1–3), and in each trial the local protocol (Arms B–D) was compared against the common protocol, Arm A (Table 1). Two languages were included in each trial (Trial 1 Danish and Swedish, Trial 2 Finnish and Swedish, and Trial 3 English and Norwegian). Speech results in the Scandcleft project at age 5 years revealed few differences related to surgical method. Consonant proficiency was assessed in terms of percentage of consonants correct (PCC), originally developed by Shriberg and Kwiatkowski (1982) and used as a severity measure based on phonetically transcribed connected speech. PCC has later been used with target consonants in-word tests to assess articulation skills in children with and without using the severity measure (Lohmander & Persson, 2008; Scherer et al., 2008; Klintö et al., 2014). Further, this modified PCC has been recommended as a standard overall measure of consonant articulation by the International Consortium for Health Outcomes Measurement (ICHOM) (Allori et al., 2017).

The only statistically significant difference in the Scandcleft project at age 5 was with the PCC score. Arm B in Trial 1, with long delay hard palate closure at age 3, showed a significantly lower PCC score than short delay hard palate closure at 12 months of age (Arm A) (Willadsen et al., 2017). Both Arms A and B had their soft palate closed at 3–4 months of age. The highest proportion of children with age-appropriate (> 90%) PCC scores were 44% (Arm D, early hard palate closure with vomer flap at 3–4 months of age and hard palate closure at 12 months). This benchmark measure (PCC > 90%) was not significantly different in any of the three trials (Hammarström et al., 2020; Persson et al., 2020; Willadsen et al., 2019). The outcome of velopharyngeal function at age 5 was based on the validated composite variable (VPC-Sum). This variable included assessments of target sounds in single words by narrow phonetic transcription on the speech characteristics of velopharyngeal incompetence (VPI); (1) passive VP symptoms; audible nasal air leakage accompanying oral pressure consonants (including nasal emission and nasal turbulence), reduced pressure on oral pressure consonants; and (2) presence of non-oral errors and active nasal fricatives. The presence of an error was decided depending on the number of consonant tokens affected (less than three = absent, three to five affected = present with a mild occurrence, and more than six affected = moderate to severe occurrence).

The final characteristic (3) hypernasality was assessed by category scale rating on nine of the single words, including high vowels in similar context edited to a string without pauses (Lohmander et al., 2017b). The summary from all included variables varied between 0 and 6 and represented the VPC-Sum. No statistically significant differences in velopharyngeal function across trials were found. The surgical method with highest prevalence of velopharyngeal competence (VPC), assessed using the VPC-Sum, had 62% (Arm B, Trial 1; long delay hard palate closure) (Lohmander et al., 2017a). A comparison across centres at age 5 in the Scandcleft project found a substantial variation in outcome of the same surgical method (Hammarström et al., 2020; Persson et al., 2020; Willadsen et al., 2019). This implied that other factors such as surgeons' learning curve (Rautio et al., 2017), skills and experience (Williams et al., 1999; Shaw & Semb, 2017) should be taken into account.

It is also important to follow the effect of time, as speech is expected to improve with increasing age (Lohmander, 2011). A few studies have reported speech outcomes at 10 years of age (Brunnegård & Lohmander, 2007; Feragen et al., 2017; Havstam et al., 2011; Lohmander et al., 2006, 2012; Morrison et al., 2022; Nyberg et al., 2014), one at 11–14 years (Grunwell et al., 2000), and one at 12 years (Sell et al., 2001). This age is often chosen as children have spent some years in school and, depending on the school system, they are often about to start secondary school, giving a higher demand on their speech. Further, this age was recommended in practice guidelines following the Eurocleft project (Shaw et al., 2001).

Besides surgical methods, timing of surgery, and surgical skills and experience, other factors such as gender,

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TABLE 1 Participating centres, languages and surgical procedures: Participants with speech recordings at age 10 years

Trial no. Recorded/total number randomized to trial	Participating centre	Language	Surgical protocol		Arm
1 ($n = 144/148$)	1[a], 2[a], 3	Danish, Swedish	Arm B Long delay hard palate closure	Arm B Lip and soft palate closure at 3–4 months. Hard palate closure at 36 months	A ($n = 72$) B ($n = 72$)
2 ($n = 125/151$)	4–6	Finnish, Swedish	Arm C Simultaneous hard and soft palate closure	Arm C Lip closure at 3–4 months. Hard and soft palate closure at 12 months	A ($n = 63$) C ($n = 62$)
3 ($n = 130/149$)	7–10[b]	Norwegian, English	Arm D Early hard palate closure with vomer flap	Arm D Lip closure at 3–4 months combined with a single-layer closure of the hard palate using a vomer flap. Soft palate closure at 12 months	A ($n = 62$) D ($n = 68$)
Common	To be carried out by all centres		Arm A Short delay in hard palate closure	Arm A Lip and soft palate closure at 3–4 months. Hard palate closure at 12 months	A ($n = 197$)

Notes: [a]All patients are operated at one centre but followed up at two centres.
[b]Centre 10 is not included at age 10; ended follow-up after 5 years of age ($n = 17$); n = total recorded/total randomized eligible at age 10 and was 399/431.

amount of speech therapy and hearing level may influence speech outcome. Two large scale studies have reported a gender difference in children with UCLP at age 5 (Butterworth et al., 2022; Willadsen et al., 2017). Girls had better articulation compared with boys (Butterworth et al., 2022; Willadsen et al., 2017) and boys had lower odds of achieving speech within the normal range (Butterworth et al., 2022). It remains to be studied if a gender difference is present in 10-year-olds born with UCLP.

Young children with cleft palate demonstrate a higher prevalence of otitis media with effusion (OME) than children without cleft palate and this trend seems to continue throughout childhood and into adolescence with a decrease in OME as children become older (Flynn et al., 2013; Kwan et al., 2011; Yang et al., 2012). While OME does not necessarily cause a hearing loss, mild fluctuating conductive hearing loss is often associated with OME (Flynn et al., 2009; Kwan et al., 2011), which could influence speech–language development (Broen et al., 1998; Schönweiler et al., 1999), and may be a predictor of poor speech (Hall et al., 2017).

The peer-reviewed evidence for the benefit of speech therapy for individuals born with CLP is uncertain (Vallino-Napoli, 2011), and is given with a wide range of intervention methods (Williams et al., 2021) without strong evidence to support the effectiveness of any specific intervention in children with cleft palate (Bessell et al., 2013). However, Sand et al. (2022) concluded that many individuals benefit from speech therapy, even though less than a third of the individuals' speech production reached the level of their peers after speech therapy. Therefore, it is important to control for the number of speech therapy visits across arms and trials.

Consonant proficiency in 10-year-olds born with UCLP

Consonant articulation outcomes have most commonly been reported with a focus on error types related to the cleft palate condition, active cleft speech characteristics (CSCs) (John et al., 2006). The focus has mainly been on active posterior/retracted oral CSCs including palatal/velar realizations of alveolar consonants and non-oral errors such as glottal articulation of oral consonants (Willadsen et al., 2017). The most common proportion of the error type, retracted oral articulation, that is, palatal or velar for dental/alveolar stops, was below 10% in 10-year-olds with UCLP. This was assessed by rating the occurrence on a five-point scale (n = 147) with very similar definitions. Later the assessments were pooled to a three-point scale: (0) normal/absent/slight, mild, (1) moderate and (2) severe/almost always (Lohmander et al., 2006; Nyberg et al., 2014) or a four-point scale: (0) normal/absent/slight, (1) mild, (2) moderate and (3) severe/almost always (Lohmander et al., 2012). Havstam et al. (2011) used an overall four-scale rating of articulation and reported a similar outcome; less than 15% (n = 54) had moderate to severe deviance including oral retracted and glottal articulation. Speech was rated on repeated standard sentences in most studies (Havstam et al., 2011; Lohmander et al., 2006; 2012; Nyberg et al., 2014). Grunwell et al. (2000) used a different approach in their cross-linguistic study (n = 131) and rated target consonants in sentences as correct, almost correct or incorrect. The target consonant /s/ was the only consonant rated incorrect in more than 5% of the possible consonants. Nyberg et al. (2014) reported that 30% (n = 69) of 10-year-olds had s-distortions, such as lateral, palatal or interdental (scale value 2–4). Thus, despite a variety of assessment methodologies, speech materials and languages included, these studies have reported a rather low prevalence of CSCs but often with long-lasting difficulties with s-distortions. Although this implies a rather high consonant proficiency, this has not been investigated in 10-year-olds.

Velopharyngeal function in 10-year-olds born with UCLP

Adequate velopharyngeal function is a main goal in the treatment of cleft palate and is closely related to the success of the primary surgery of the palate. Overall perceptual ratings have been found to be representative of the listeners' overall impression of degree of velopharyngeal impairment (e.g., Karnell & Van Demark, 1986; McWilliams et al., 1981) and a sum of assessed speech symptoms of velopharyngeal insufficiency (composite score) reliable (Pereira et al., 2021). Further, the composite scores correlate well with an overall perceptual rating of velopharyngeal function (Dotevall et al., 2002; Lohmander et al., 2009). As earlier described the validated composite variable (VPC-Sum) in the Scandcleft project was based on assessments of target sounds in single words and a scale rating of hypernasality (Lohmander et al., 2017b). VPC rating (VPC-Rate) on the other hand, was based on an overall rating of perceived velopharyngeal function of connected speech along a three-point ordinal scale.

Havstam et al. (2011) applied an overall rating procedure on a four-point scale (competent, marginally incompetent, mildly moderately incompetent and severely incompetent) on speech material based on standard sentences. They found that almost half the group with cleft palate (+/– lip) had mildly or moderately incompetent VP function at 10 years of age. In comparison, only 7% had this difficulty in the UCLP group investigated by Nyberg et al. (2014).

However, in that study, an estimation of primary VPI was 50%. That is, when adding the number of children who had received secondary velopharyngeal surgery to improve speech (43%) to the group with perceived VPI (7%) at age 10 years. Also, primary VPI in approximately half the UCLP group was reported by Lohmander et al. (2006) with mild to moderate VPI found in 38%, and in addition 15% had received velopharyngeal flap surgery, which is a type of velopharyngeal surgery. In summary, primary VPC seems to have been obtained in about half of the children treated for UCLP.

The evidence base for the best possible surgical procedure and timing of surgery for children born with UCLP is still weak. To date, information on speech outcome in school-aged children around 10 years of age treated for UCLP report rather poor group results (Havstam et al., 2011; Lohmander et al., 2006; Nyberg et al., 2014) and include few individuals. Therefore, further information on speech outcome after primary surgical repair in this age group is warranted and would further clarify the relationship between surgical protocol and speech outcome.

The overall aim of the study was to describe and compare speech proficiency outcomes at 10 years of age in the Scandcleft project after different surgical protocols of the cleft palate. The following research questions were posed:

- What is consonant proficiency (PCC score) like at age 10 years and is it influenced by surgical protocol (Arm A, B, C or D), velopharyngeal competence at the same age, or gender?
- What is Velopharyngeal competence like at age 10 years and is it influenced by surgical protocol (Arm A, B, C or D) or secondary pharyngeal surgeries?

METHOD

Participants

A total of 448 children born with non-syndromic complete UCLP were recruited during a 9-year period (1997–2006) to the Scandcleft project. To be included in the trials children should be of Caucasian origin, have a maximum width of 5 mm of a possible soft tissue bridge (Simonart's band) and the native language of the country of residence had to be spoken by at least one caregiver. For a more thorough description of the recruitment, randomization and participant flow, see Semb et al. (2017). Outcomes from the main outcome age at 5 years have been reported (e.g., Hammarström et al., 2020; Lohmander et al., 2017a; Persson et al., 2020; Semb et al., 2017; Willadsen et al., 2017, 2019) and follow-up data from age 10 have been collected. One British centre in Trial 3 ($n = 17$) did not continue follow-up beyond 5 years of age due to lack of resources and ethical approval. Accordingly, this study reports findings from nine centres with 431 eligible participants randomized to the trial. Speech recordings at age 10 were available from 399 of 431 participants (92.6%; 140 girls, 259 boys) with variation between trials (Trial 1: 97.3%; Trial 2: 82.8%; Trial 3: 87.2%). They were recorded at a mean age of 10.09 years, standard deviation (SD) = 0.26. Thus, 32 recordings were missing. Either the children did not attend the 10-year visit ($n = 18$), did not cooperate ($n = 2$), the recording equipment did not work ($n = 2$) or they were not recorded at one centre due to lack of resources ($n = 10$).

Three parallel clinical trials evaluated the outcome of a standard surgical protocol (Arm A) against three different surgical protocols (Arms B–D) (Table 1). Trial 1 compared speech outcome in relation to timing of hard palate closure; Arm A (short delay in hard palate closure at 12 months of age) and Arm B (long delay in hard palate closure at 3 years of age). In both arms the soft palate was repaired at 3–4 months of age. Trial 2 compared a two-stage procedure with a one-stage procedure; Arm A (short delay in hard palate closure at 12 months of age and soft palate closure at 3–4 months of age) and Arm C (simultaneous hard and soft palate closure at 12 months of age). Trial 3 compared the sequence in two-stage procedures; Arm A (short delay in hard palate closure at 12 months of age and soft palate closure at 3–4 months of age) and Arm D (early hard palate closure with vomer flap at 3–4 months of age and soft palate closure at 12 months of age). All children were randomized to one of the two surgical procedures performed at the child's cleft centre. For a more detailed description of the surgical procedures, see Rautio et al. (2017).

Ethical approval was obtained in each participating country. The study has trial registration ISRCTN29942826.

Secondary pharyngeal surgeries

By 10 years of age, a total of 118 children (29.6%) had received secondary pharyngeal surgery. A total of 33 of these had their surgery before age 5 years. Table 2 provides an overview of the distribution of number of children having received secondary pharyngeal surgeries (%) and statistical analysis by trial and arm. Decision on secondary pharyngeal surgery were based on the local cleft centres' regular protocols. Around half of the procedures were velopharyngeal flaps and around one quarter was the Furlow procedure. The remaining included other procedures or combinations of procedures. There was no statistically significant difference between arms in any of the three trials.

TABLE 2 Number of children (%) having received VPI surgery and number of speech therapy visits per child in each trial and arm by 10 years of age

	Trial 1		Trial 2		Trial 3	
Arm	**A**	**B**	**A**	**C**	**A**	**D**
Number (%) of children having secondary VPI surgery	17 (23.6)	14 (19.4)	19 (30.2)	17 (27.4)	27 (43.5)	21 (30.9)
Difference between arms	$z = 0.61$; $p = 0.543$		$z = 0.34$; $p = 0.735$		$z = 1.49$; $p = 0.135$	
95% CI	Lower: −9.24 Upper: 17.58		Lower: −13.13 Upper: 18.61		Lower: −3.85 Upper: 29.19	
Mean (SD) number of speech therapy visits	29.7 (33.6) $n = 69$	63.4 (69.4) $n = 71$	37.4 (68.3) $n = 51$	31.2 (37.1) $n = 49$	53.0 (52.6) $n = 61$	35.3 (44.6) $n = 65$
Difference between arms	$t = -3.67$; $p < 0.001$		$t = 0.57$; $p = .569$		$t = 2.03$; $p = 0.045$	
95% CI	Lower: −51.89 Upper: −15.50		Lower: −15.51 Upper: 28.01		Lower: 0.40 Upper: 34.92	

Speech therapy visits

Total number of speech therapy visits up to 10 years of age including the routine visits at the cleft clinic, follow-up visits, and speech and language intervention visits are summarized and calculated in Table 2. Information regarding type of intervention was not available. In Trial 1, Arm B had a statistically significant higher number of speech therapy visits (mean = 63.4) compared with Arm A (mean = 29.7), $t = -3.67$; $p < 0.001$. There were no other statistically significant differences between arms.

Hearing

Pure tone audiometry (PTA) was performed in 275 out of 395 (69%) children at age 10; in Trial 1 87%, in Trial 2 48% and in Trial 3 69%. Average hearing thresholds (PTA) on four frequencies (500, 1000, 2000 and 4000 Hz) were calculated for the best ear. As pure tone audiometry was missing in 120 children, only descriptive data are reported. In Trial 1, one (1.5%) out of the 63 children had mild hearing loss (21–40 dBHL) and one (1.5%) had moderate hearing loss in Arm A. In Arm B seven (11%) out of 62 had mild hearing loss. In Trial 2, which only included half of the group, one (3.6%) out of the 28 children had mild hearing loss in Arm A, and one (3.2%) out of the 31 tested in Arm C had mild hearing loss. In Trial 3, where approximately two thirds of the group were included, five (11%) out of 45 in Arm A had a mild hearing loss compared with two (4.6%) out of 43 children in Arm D.

Procedure

Speech material

Consonant articulation was assessed at age 10 by means of the same single-word naming test developed for each one of the five included languages for possible use at age 3 years. To minimize the influence of language background, we included a restricted speech material of target consonants that were as similar as possible across the languages. The language-specific target words were selected based on a common set of phonetic principles, for example, the target consonant should be in strong position and should be followed by a vowel, and the words should not include nasals or consonant clusters (see Lohmander et al., 2009, for a full description). In addition, target consonants should be vulnerable to the cleft condition and therefore mainly obstruents were included (high-pressure sounds) (e.g., Hutters & Henningsson, 2004).

The restricted single-word naming test contained 33 words, with one of the target consonants /p t k b d g f s v n/ in each word. Each target consonant was assessed three times in different words in word-initial, stressed position, except for /s/ that was also assessed three times in word-final position as this is established earlier than the initial position in younger children (Lohmander et al., 2009). Finnish was an exception as the language does not include / b d g f /. Therefore, the Finnish naming test included six examples of /p t k/ and a total of 30 target words. The other consonants were the same as for the other languages (Lohmander et al., 2009). For the single-word naming test, reading was used; if this failed, a hierarchy of picture naming, semantic prompting or word repetition was used. The examiner repeated the target words after the child to identify the words and to add a native adult production of the target sound to compare the child's production with.

The single-word naming tests were later used for transcription of target consonant production, including passive VP symptoms (nasal emission, velopharyngeal friction sounds and weak pressure consonants).

To rule out the possible impact of language background as much as possible when assessing hypernasality (Lohmander et al., 2017a) the first nine words in the single-word naming test were edited to a string in wav.format for each child. These nine words contained high or semi-high vowels in a similar phonetic context across languages.

A 2-min sample of continuous speech was used for perceptual rating of velopharyngeal function (VPC-Rate) and rated on conversational and rote speech. Where this was missing retelling of the Bus Story was used (Renfrew, 1997).

Data recording and editing

All participants were seen by two speech and language pathologists/therapists (SLP/T) to each cleft centre as close as possible to their 10th birthday. Each participant were video recorded using a super-VHS or digital video camera with an external microphone of excellent quality (e.g., the video camera SonyDCRTRV30E/similar and a microphone Sony ECMMS957, Londerzeel, Belgium). One of the SLP/Ts, the examiner, sat opposite the child with the camera operator just behind with the camera lens directed towards the child's face. Two external microphones (one for the audio recording and video recordings, respectively) were placed approximately 40 cm from the child. The test pictures with written text were presented in such a way that the child looked straight into the camera.

All video recordings were centrally edited before the assessments by four assistants, specifically trained for the project. Each video recording of the single word test, including the examiner's repetition of the target words, was saved as a mp4 file. If the video recording failed the audio recording was used for analysis. A second file including 2 min of continuous speech was edited and saved as an mp4 file. The nine first words were edited into a nine-word-string without pauses between the words, that is, the clip was edited as close as possible to the beginning and end of each word before they were merged and saved as audio wav.files.

Training of perceptual assessment of speech

All but four of the SLP/Ts ($n = 16$) had earlier assessed the Scandcleft 5-year data. These four SLP/Ts, one each from Denmark, Finland, Norway and the UK were experienced SLP/Ts working in cleft clinics. They have participated in the speech Scandcleft group since the 5-year assessment. During the development of the trial and before the 5-year analyses, extensive training of phonetic transcription and rating of velopharyngeal function and hypernasality had taken place every second year for 2–3 days (Lohmander et al., 2017a; Willadsen et al., 2017). However, no common training had taken place since the 5-year assessments. Before the 10-year assessment began the SLP/T group had online training and a common training session at the venue, which took place the day before the assessments started. Rules for transcription and video examples of different error categories, such as passive errors, active oral errors and active non-oral errors but also language-specific errors and developmental speech characteristics, were trained and discussed in plenum. The video examples used had previously obtained complete interrater agreement during training/calibration and were used as anchors in the discussions. In addition, examples from different error categories were rated and discussed in common. Written rules for transcription, earlier used at the 5-year assessment, were discussed and agreed. Further, training on rating of the velopharyngeal function (VPC-Rate) and hypernasality was performed in plenum. The group listened to anchors of each scalar points, earlier agreed on by the group. Thereafter each rater subsequently undertook individual ratings of recordings followed by a common discussion.

Perceptual assessment

The SLP/Ts independently viewed the video recordings on a laptop, wearing high-quality headphones (Creative Aurvana Live, Creative Technology Ltd, Singapore/similar). All naming tests were assessed by two SLP/Ts blinded to randomization of the children. For all participating countries/languages, except for Finland, at least two centres were part of the study. Therefore, the SLP/Ts phonetically transcribed speech samples of their own language, but from a different centre—except in two cases: (1) the Finnish samples were transcribed by a Finnish SLP/T and two Swedish SLP/Ts (who assessed 62 and 20 recordings, respectively). The transcriptions by the Finnish SLP/T were used in all analyses. The Swedish transcriptions were used for interrater agreement. (2) The English samples were transcribed by two English-speaking SLP/Ts, one from the same centre as the participants. However, transcriptions from the external SLP/T were used for analyses. The SLP/Ts transcribed the children's realization of target consonants using narrow phonetic transcription according to IPA and extIPA.

Hypernasality was rated as a two-step procedure, according to the procedure at the 5-year assessment (Lohmander et al., 2017a). The SLP/Ts could listen to the nine-word string as many times as needed. In the first step the SLP/T decided if resonance was within normal limits. If not, the SLP/T decided if hypernasality was mild, moderate or severe. Thus, a two-step four-point ordinal scale was used. The overall velopharyngeal function based on continuous speech was rated on a three-point scale (VPC-Rate); competent, marginally incompetent or incompetent (Lohmander et al., 2017b). For both variables each recording was rated by three SLP/Ts, two speaking the same language as the child but from a different centre and one SLP/T speaking one of the other languages. There were four exceptions, one of the SLP/Ts speaking the same language as the child came from the same centre in one Danish, one Swedish and one British centre and both Finnish SLP/Ts came from the same centre as the children. Outcome was classified by a majority decision if at least two of the three SLP/Ts agreed. If all three disagreed, the middle value was chosen.

Analysis

Consonant proficiency

In the present study PCC was calculated for each participant as the number of correctly produced target consonants out of all target consonants produced as described in the introduction. This means that each target consonant was scored as correct or incorrect based on the phonetic transcription. Any deviance according to articulation place or manner was scored as incorrect. If a target consonant was produced correctly, apart from passive VPI symptom, it was scored as correct. As the s-sound can be a remaining difficulty at age 10 and had a high frequency in the test ($n = 6$), also percentage consonants correct without /s/ (PCC-s) was calculated for each participant to assess the consonant proficiency of the remaining target consonants. In contrast to the PCC score, all s-targets produced (six out of 33 target consonants) were scored as correct in this measure. The PCC scores were compared with normative data from 60 Swedish-speaking 10-year-olds without a cleft palate, who named the same consonants in same or similar single words as in the Scandcleft project (Lohmander et al., 2017c). The latter group had an average PCC of 97% (SD 6), thus ≤ 1 SD was 91% and ≤ 2 SD was 85%. Normative PCC data were not available for the Scandcleft languages other than Swedish. Thus, the Swedish standard of ≥ 91 was taken as the standard for age-appropriate PCC. As the present study focused on consonant proficiency, only s-distortions were presented in addition to the PCC score.

Velopharyngeal competence (VPC)

Velopharyngeal function was measured using the composite score VPC-Sum earlier described. For a more detailed description, see Lohmander et al. (2017a, 2017b). In the present study the VPC-Sum was presented as a dichotomized value according to Lohmander et al. (2017b). VPC-Sum score 0–1 was considered as VPC and score 2–6 as VPI, including both marginal incompetence and incompetence. Thus, VPC can include one of the symptoms scored as VPC-Sum 1 (mild hypernasality, 3–5 affected consonants of either VP symptoms or active non-oral errors).

To study the VPC as a consequence of the primary surgery, the primary VPC/VPI were calculated. Primary VPC included score 0–1 on the VPC-Sum and no secondary VP surgery, while primary VPI included VPC-Sum score 2–6 and/or VP surgery.

Inter- and intra-rater reliability

Inter-transcriber reliability for correct or incorrect realization of all target consonants based on phonetic transcription was assessed as point-by-point agreement between the pairs of SLP/Ts assessing the same recordings, altogether 12 pairs of raters. All differences between correct/incorrect were counted as disagreements. Two pairs had a mean agreement between 86.6% and 88%. The other 10 pairs had a mean agreement > 90%.

For assessment of intra-transcriber reliability, 20% of the recordings randomly distributed among the previous assessments were duplicated. All SLP/Ts had a mean agreement > 92%. Therefore, both inter- and intra-agreement on the articulation scores were considered acceptable.

In 47% of the 394 ratings of the VP function along a three-point ordinal scale (VPC-Rate) all three SLP/Ts agreed on their own ratings, in 49% of the recordings two of the SLP/Ts agreed, and in 4% of the ratings all three SLP/Ts disagreed. Thus, in 96% of the ratings a majority decision was possible. The exact intra-rater agreement across each one of the SLP/T varied between 66% and 100%, with a mean of 84%.

In 49% of the 394 ratings of hypernasality on a four-point ordinal scale, all three SLP/Ts agreed on their ratings, in 46% of the ratings two of the SLP/Ts agreed and in 5% of the ratings all three SLP/Ts disagreed. Thus, in 95% of the ratings a majority decision was possible. The exact intra-rater agreement across the SLP/Ts varied between 58% and 100%, with a mean of 76%.

Statistical analysis

Descriptive analyses and a generalized linear mixed logistic model were used. PCC scores derived from correct and incorrect consonant realizations of target words in each child for which all demographic variables were available ($n = 387$) were used as the dependent variable. This form of regression is appropriate to use when variables are not evenly balanced across subjects. In the regression analysis we use the 'lme4' (Bates et al., 2015) and 'ordinal' (Christensen, 2019) packages in the R statistics environment (R Core Team, 2019). Initial modelling was performed on a trial-wise basis, that is, three separate models for each of the three trials were prepared. This approach was discontinued after comparison of a combined model including all trials (1–3) and arms (A–D) showed that Arm A did not significantly differ between trials ($\chi^2_{(2)} = 2.508$, $p = 0.285$), which supported the combination of the data from all trials, that is, from all nine centre locations. Combining the trials was also in the interests of statistical power and this in turn supported interpretation of factors such as gender. The combined data from all centres were also based on PCC data in the whole group.

RESULTS

Consonant proficiency

Based on the phonetic transcription of the target consonants in the single-word test PCC was calculated for all included consonants (PCC) and without /s/ (PCC-s) (Figure 1). In comparison with normative data on 10-year-olds without a cleft, the following percentage of children

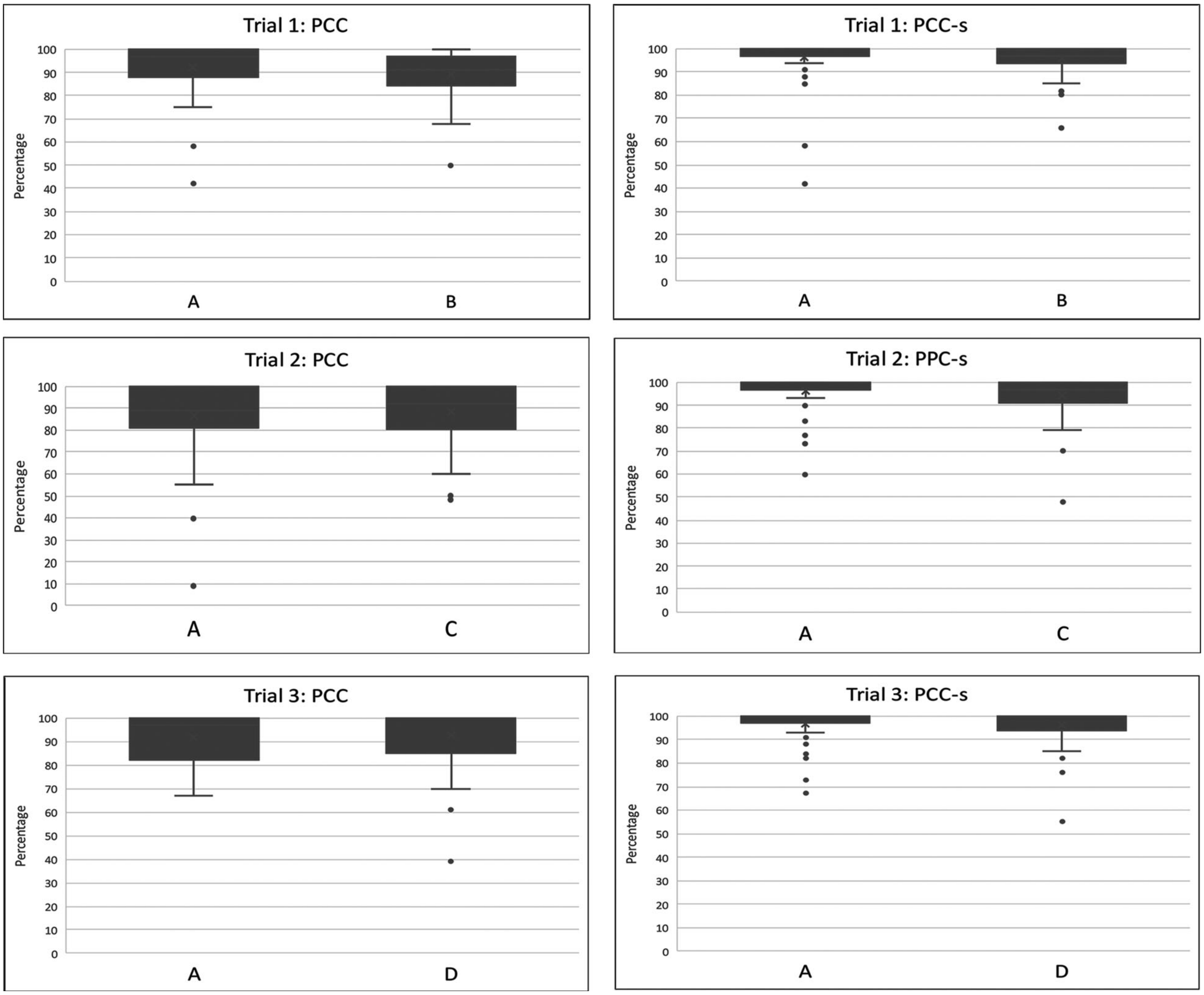

FIGURE 1 Box plot graphs showing the PCC and the PCC-s median (mean *x*), quartiles 1 and 3 for the three Scandcleft trials and arms: Arm A was common in all trials with soft palate repair at 3–4 months and a short delay in hard palate closure at 12 months; Arm B was soft palate repair at 3–4 months and a long delay in hard palate closure at 36 months; Arm C was simultaneous hard and soft palate closure at 12 months; and Arm D was early hard palate closure with vomer flap at 3–4 months and soft palate closure at 12 months. Minimum and maximum values are shown at the end of whiskers and outliers as circles.

was considered to have an age-equivalent PCC: Trial 1, Arm A (short delay in hard palate closure): 73%, Arm B (long delay in hard palate closure): 62%; Trial 2, Arm A: 50%, Arm C (simultaneous hard and soft palate closure): 60%; Trial 3, Arm A: 65%, Arm D (early hard palate closure with vomer flap): 69%. Further, a proportion of children had extensive difficulties with a PCC score < 2 SD of the mean: Trial 1, Arm A: 12%, Arm B: 24%; Trial 2, Arm A: 39%, Arm C: 21%; Trial 3, Arm A: 26%, Arm D: 17%. As can be seen in Figure 1, s-distortions were common. The remaining errors with an impact on PCC consisted of oral (retracted to palatal/velar/uvular place of articulation) and non-oral CSCs and, developmental speech characteristics (DSC) (Willadsen et al., 2017), which are not detailed in the present article.

Influence on PCC by surgical protocol, VPC and gender

The selected regression model (Table 3) shows that PCC score (consonant proficiency) of female participants were significantly better than males when analysed across all trials. A high VPC-Sum (indicating poor velopharyngeal function) was significantly associated with low PCC score. The PCC score from the participants in Arms B (long delay in hard palate closure) were significantly poorer than Arm A (short delay in hard palate closure), and Arm C (simultaneous hard and soft palate closure) were significantly better than Arm A (short delay in hard palate closure). There was no significant difference in PCC score between Arms A and D (early hard palate closure with vomer flap).

TABLE 3 Summary of fixed effects in the combined trial mixed-logit model

Predictors	Coefficient	SE	Z	p
Gender: female	0.561	0.203	2.766	0.005**
VPI surgery	0.165	0.212	−0.776	0.437
VPC sum	−0.435	0.084	−5.163	0.001***
VPC rate	−0.31	0.177	−1.747	0.081
Arm B	−0.828	0.306	−2.709	0.007**
Arm C	0.734	0.337	2.177	0.029*
Arm D	0.247	0.341	0.725	0.468
Trial 2	−0.874	0.51	−1.714	0.086
Trial 3	−0.389	0.531	−0.734	0.463

Notes: Predictors are Gender (reference: Male); VPI surgery (reference: No corrective surgery); VPC-Sum; VPC-Rate; Arm (reference: A); and Trial (reference: Trial 1). Trial is included as a validation of our combined analysis.

*$p < 0.05$;

**$p < 0.01$;

***$p < 0.001$.

Similarly, there were no significant differences between the trials, which is in part validation of our decision to analyse the combined data from all trials.

Velopharyngeal function

Three different descriptive measures of VPC were included as well as the proportion of participants having VP surgery; primary competent velopharyngeal function (Figure 2a), VPC-Sum (Figure 2b) and VPC-Rate (Figure 2c). The primary competent velopharyngeal function, defined as a VPC-Sum 0–1 and no VP surgery, varied between 46.8% (Trial 2, Arm C and Trial 3, Arm A) and 68.1% (Trial 1, Arm A) (Figure 2a). VPC-Sum (Figure 2b) including a VPC-Sum 0–1 and performed VP surgeries varied between 58.1% (Trial 2, Arm C) and 83.3% (Trial 1, Arm B). The third measure was based on ratings of continuous speech (VPC-Rate). These ratings of competent velopharyngeal function (Figure 2c) varied between 38.3% (Trial 2, Arm C) and 64.6% (Trial 3, Arm D).

Influence on VPC-Sum by surgical protocol or secondary pharyngeal surgeries

As VPC-Sum reflected velopharyngeal function and was a significant predictor of consonant proficiency, we modelled this composite score as a dependent variable. The ordinal mixed-effects regression model was therefore based on data from each child in the whole group ($n = 393$). The model (Table 4) showed that the VPC-Sum of children in Arm C (simultaneous hard and soft palate closure) were significantly worse than participants in Arm A (short delay in hard palate closure). Also, the main effect of VP surgery was significant, indicating a strong relationship between participants who had undergone secondary pharyngeal surgery (VP surgery) and participants with higher VPC-Sum, that is, with poorer velopharyngeal function. However, these findings should be interpreted with caution as the model is based on a limited number of data, and the distribution of VPC-Sums shows a preponderance of low scores.

DISCUSSION

The purpose of the study was to present and compare speech outcome at 10 years of age in children treated for UCLP according to four different surgical protocols in the Scandcleft project. Only 50–73% of children were found to have age-appropriate consonant proficiency in terms of PCC score on target consonants in single words, and 12–39% had scores at least 2 SD below, compared with published Swedish reference data (Lohmander et al., 2017c). Despite different assessment methods and different type of speech materials this level is comparable with the levels of consonant proficiency reported in other studies of 10–14-year-olds treated for UCLP; 65% (Havstam et al., 2011), 70% (Nyberg et al., 2014), 68% 12 years of age (Sell et al., 2001), and 75% (Brunnegård & Lohmander, 2007). Lohmander et al. (2012) found that 90% had no typical cleft speech errors but s-distortions were not evaluated. Grunwell et al. (2000) concluded that /s/ was the most affected phoneme at this age. Accordingly, when the s-sound was excluded in the present study the PCC scores were markedly increased, particularly in Arm A across the trials. Distortions of /s/ have often been referred to as minor errors. However, the consequences might be troublesome as highlighted by Nyberg and Havstam (2016), who found that 10-year-old

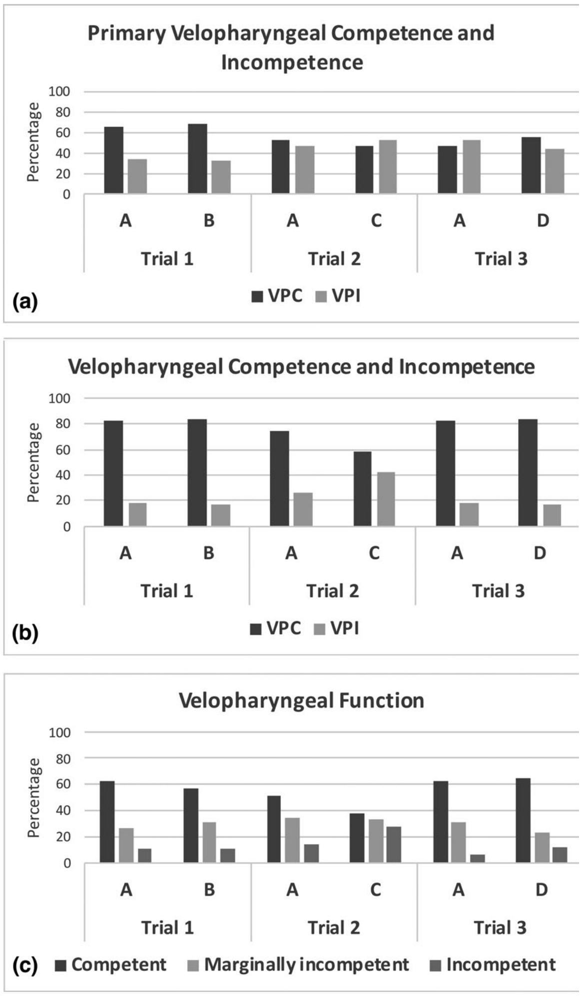

FIGURE 2 Three descriptive measures of velopharyngeal function, competence (VPC) or incompetence (VPI) and the proportion of participants in each Scandcleft trial (1–3) and arm (surgical protocol). Arm A was common in all trials with soft palate repair at 3–4 months and a short delay in hard palate closure at 12 months; Arm B was soft palate repair at 3–4 months and a long delay in hard palate closure at 36 months; Arm C was simultaneous hard and soft palate closure at 12 months; and Arm D was early hard palate closure with a vomer flap at 3–4 months and soft palate closure at 12 months. Velopharyngeal function was reported as (a) primary VPC, which includes the proportion of participants with VPC-Sum 0–1 and no VPI surgery, and primary VPI, which includes the proportion of participants with VPC-Sum 2–6 and/or VPI surgery; (b) proportion of participants with VPC based on a dichotomized VPC-Sum including value 0–1 and VPI including value 2–6; and (c) showing the proportion of participants with competent, marginally incompetent and incompetent velopharyngeal function in terms of VPC-Rate, that is, rating on a three-point ordinal scale.

peers were sensitive to distortions of the s-articulation. As this is a frequent consonant in many languages this is an error that clinicians should focus on when appropriate. A competent velopharyngeal function is an important prerequisite for good articulation development (Harding & Grunwell, 1998). This was supported by the overall data across the trials, showing that a high VPC-Sum (i.e., VPI) indicated a low PCC (poor consonant articulation). This indicates that early surgical management of VPI could be beneficial for the consonant proficiency, that is, preventive of non-oral errors. However, it cannot be ruled out that this relationship between the VPC-Sum and PCC was affected by the fact that non-oral errors are part of the VPC-Sum. Notwithstanding, non-oral errors were only one of three variables comprising the VPC-Sum and three or more non-oral errors were only found in 8% of the children.

In general, girls had significantly better PCC scores than boys in the present study. Thus, the findings from age 5 (Butterworth et al., 2022; Willadsen et al., 2017) was still present at age 10. However, the same pattern with girls having a better PCC score than boys has also been reported in typically developing children speaking British English at age 5 (Dodd et al., 2003) and in children with persistent speech sound disorder at age 8 (Wren et al., 2016). Thus, it has been hypothesized that gender difference might correspond to typical development in boys (Butterworth et al., 2022; Willadsen et al., 2017). Whether the gender difference among children with UCLP is a common pattern at 10 years of age needs to be confirmed in future studies.

The overall prevalence of children with VPC at age 10 years seems rather low. The occurrence of VPC in terms of VPC-Sum in the present study varied between 58% (Trial 2 Arm C) and 83% (Trial 1 Arm B; Trial 3 Arm D). The occurrence of VPC in terms of ordinal rating (VPC-Rate) on continuous speech revealed a variation in VPC between 38% (Trial 2 Arm C) and 65% (Trial 3 Arm D). Although different levels of percentage were observed between the methods, the pattern across trials and arms were the same (Figure 2). The occurrence of VPC in terms of VPC-Rate was comparable to what has previously been reported at 10 years of age (55%) (Havstam et al., 2011) but was lower compared with outcome (93%) reported by Nyberg et al. (2014), where, however, 43% had received VP surgery. Thus, outcome data of velopharyngeal function at age 10 was influenced by VP surgery. Therefore, primary VPC, the perceptual assessment of velopharyngeal function without influence of secondary VP surgery was calculated. In the present study primary VPC was defined as VPC-Sum 0–1 and no VP surgery. As expected, the proportion of children with primary VPC was lower than the percentage with VPC-Sum 0–1 and varied between 47% (Trial 2 Arm C, Trial 3 Arm A) and 68% (Trial 1 Arm B). These figures are comparable with the primary VPC of 50% at age 10 (Nyberg et al., 2014) and 68% at age 12 (Sell et al., 2001). There was also a quite large variation in number of performed VP surgery in the different Arms, but no statistically significant difference in any of the trials. However, Trial 3 had the highest proportion of participants who had received VP

TABLE 4 Summary of fixed effects in the cumulative link mixed model ($n = 393$) with VPC-Sum as the dependent variable

Predictors	Coefficient	SE	Z	p
Gender	−0.419	0.219	−1.907	0.056
Arm B	0.159	0.274	0.582	0.561
Arm C	1.074	0.274	3.759	< 0.001***
Arm D	−0.104	0.303	−0.346	0.729
VPI surgery	0.776	0.216	3.594	< 0.001***

Note: Predictors are Gender (reference: male); Arm (reference: A); and VPI surgery (reference: No corrective surgery).

surgery followed by Trial 2, giving an impact on the burden of care. Nevertheless, our statistical calculation across trials reveal that VP surgery performed indicate a high VPC-Sum. In other words, many of the children still had difficulties with the VP function after VP surgery. This is in line with an earlier study reporting that most individuals showed an improved VP function, but not VPC (Liedman-Boshko et al., 2005). These authors also noticed that the later the speech assessment after VP surgery, the better the speech. This factor was not included in the analysis in the present study. Thus, differences in and impact of VPI surgeries need to be understood in terms of different local protocols and practice (Rautio et al., 2017).

The mean number of speech therapy visits was high across the trials but with large variation. A statistically significant difference between arms was found in Trials 1 and 3 with the highest number of speech therapy visits in Arm B (Trial 1) and Arm A (Trial 3). These two arms had different types of main difficulties. Arm B had a high number of active oral CSCs at age 5 (Willadsen et al., 2017) and a lower PCC at age 10 compared with Arm A, whereas Arm A in Trial 3 had the highest number of VPI surgeries. We lack information on type of speech intervention given, but in both trials the SLPs seems to have spent most resources on the participants with speech difficulties of different kinds. Children had had as many as over 350 speech therapy visits. As this give a very high burden of care the effectiveness of the speech therapy needs to be studied in more detail.

Strengths and limitations

The strengths in the present study were the use of comparable speech material across the different languages, high quality audio-video recordings of the speech material, valid speech variables and speech assessment performed by trained SLP/Ts, speaking the same language as the speaker, but not from same centre. The agreement regarding transcription was very good, whereas it varied between poor and acceptable for hypernasality and VPC-Rate. Three SLP/Ts rated each speech samples for hypernasality and VPC-Rate. Outcome was based on majority agreement, which could be reached in 95–96% of the ratings. An advantage with three raters is that an outlying decision do not affect the outcome as long as not all three raters disagree. However, in less than half of the ratings all three raters agreed, which preferably could have been higher. It is well known that raters have their own internal standards (Kreiman et al., 1993; Keuning et al., 2004). In this trial the SLP/Ts were all experienced but came from five different countries with different concepts of assessment. Thus, it was a challenge to calibrate the SLP/Ts. Although, they have trained with the Scandcleft methodology, more training had been preferable but was not possible. To overcome these difficulties with agreement on scale rating two assessment methods of the VP function were used and hypernasality was included in the composite score VPC-Sum to make the outcome more robust. The agreement level as well as the influence by non-oral errors might impact the VPC-Sum in this study. Thus, the reported significant effect of VPC-Sum on PCC might be somewhat misleading.

The number of speech therapy visits was based on a form including different aspects of speech therapy, such as routine assessments, reviews, speech treatment and type of treatment. Unfortunately, the information in the collected forms was incomplete and inconsistent. It was therefore decided to use only total number of visits as an indication of burden of care.

It has been argued that the influence of an otitis media with effusion related hearing loss on speech development is present at early ages (Broen et al., 1998; Lohmander et al., 2021), and may be at risk for delayed speech and language development (Hall et al., 2017; Schönweiler et al., 1999). Unfortunately, the hearing data in the present study were incomplete and could not be included in the analysis. Efforts to improve the documentation of hearing longitudinally is strongly suggested.

The regression analysis revealed the effect from one variable, everything else being equal, which in reality may not be the case. The treatment result might be influenced by other variables than the primary treatment or by a combination of variables such as timing from VP surgery to follow-up (Liedman-Boshko et al., 2005), additional language disabilities, speech therapy dose and timing, and hearing, which preferably should be taken into account in order to see the whole picture. Furthermore, we have not counted for the possible effect of the surgeons' learning curve and surgical skills and experience (Rautio et al., 2017; Shaw & Semb, 2017). A longitudinal analysis of change in speech from 5 to 10 years of age including aspects of speech errors and an in-depth analysis of impact of speech therapy is warranted.

SUMMARY AND CONCLUSIONS

The mean levels of consonant proficiency in terms of PCC varied between 86% and 92%. Only 50–73% had a consonant proficiency at 10 years of age in level with peers. The VPC based on the composite score VPC-Sum varied between 58% and 83%, and the primary VPC (excluding VPI surgeries) between 47% and 68%. There were few significant predictors. Girls performed better throughout. Arm B (long delay in hard palate closure) was found to indicate lower PCC and Arm C (simultaneous hard and soft palate closure) higher PCC. At the same time, the proportion of participants with primary VPC was highest in Arm B (68%) and lowest in Arm C (47%). Thus, the different protocols had their pros and cons and there is no obvious evidence to recommend any of the protocol as superior. Other factors than primary surgical method, such as time after VP surgery, surgical experience, hearing level, language difficulties, speech therapy, or perhaps a combination of factors that may influence speech proficiency, should be thoroughly reviewed and considered in future studies.

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CONFLICT OF INTEREST

The authors declare that there is no conflict of interest.

DATA AVAILABILITY STATEMENT

This study is ongoing and the data will be used for additional purposes within the project. Data are therefore currently not available.

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REFERENCES

Allori, A.C., Kelley, T., Meara, J.G., Albert, A., Bonanthaya, K., Chapman, K., et al. (2017) A standard set of outcome measures for the comprehensive appraisal of cleft care. *Cleft Palate–Craniofacial Journal*, 54, 540–554. Available from: https://doi.org/10.1597/15-292

Bates, D., Maechler, M., Bolker, B. & Walker, S. (2015) Fitting linear mixed-effects models using lme4. *Journal of Statistical Software*, 67, 1–48. Available from: https://doi.org/10.18637/jss.v067.i01

Bessell, A., Sell, D., Whiting, P., Roulstone, S., Albery, L., Persson, M., et al. (2013) Speech and language therapy interventions for children with cleft palate: a systematic review. *Cleft Palate–Craniofacial Journal*, 50, 1–17. Available from: https://doi.org/10.1597/11-202

Britton, L., Albery, L., Bowden, M., Harding-Bell, A., Phippen, G. & Sell, D. (2014) A cross-sectional cohort study of speech in five-year-olds with cleft palate ± lip to support development of national audit standards: benchmarking speech standards in the United Kingdom. *Cleft Palate–Craniofacial Journal*, 51, 431–451. Available from: https://doi.org/10.1597/13-121

Broen, P.A., Moller, K.T., Carlstrom, J., Doyle, S.S., Devers, M. & Keenan, K.M. (1996) Comparison of the hearing histories of children with and without cleft palate. *Cleft Palate–Craniofacial Journal*, 33, 127–133. Available from: https://doi.org/10.1597/1545-1569_1996_033_0127_cothho_2.3.co_2

Broen, P.A., Devers, M.C., Doyle, S.S., Prouty, J.M., Moller, K.T. (1998) Acquisition of linguistic and cognitive skills by children with cleft palate. *Journal of Speech, Language and Hearing Research*, 41(3):676–687. Available from: https://doi.org/10.1044/jslhr.4103.676

Brunnegård, K. & Lohmander, A. (2007) A cross-sectional study of speech in 10-year-old children with cleft palate: results and issues of rater reliability. *Cleft Palate–Craniofacial Journal*, 44, 33–44. Available from: https://doi.org/10.1597/05-164

Butterworth, S., Fitzsimons, K.J., Medina, J., Britton, L., Van Eeden, S., Wahedally, H., et al. (2022) Investigating the impact of patient-related factors on speech outcomes at 5 years of age in children with a cleft palate. *Cleft Palate–Craniofacial Journal*, -Epub ahead of publication. Available from: https://doi.org/10.1177/10556656221110094

Christensen, R.H.B. (2019) R package version 12-10. Available at: https://CRAN.R-project.org/package=ordinal

Dodd, B., Holm, A., Hua, Z. & Crosbie, S. (2003) Phonological development: a normative study of British English-speaking children. *Clinical Linguistics & Phonetics*, 17, 617–643. Available from: https://doi.org/10.1080/0269920031000111348

Dotevall, H., Lohmander-Agerskov, A., Ejnell, H. & Bake, B. (2002) Perceptual evaluation of speech and velopharyngeal function in children with and without cleft palate and the relationship to nasal airflow patterns. *Cleft Palate–Craniofacial Journal*, 39, 409–424. Available from: https://doi.org/10.1597/1545-1569_2002_039_0409_peosav_2.0.co_2

Feragen, K.B., Aukner, R., Særvold, T.K. & Hide, Ø. (2017) Speech, language, and reading skills in 10-year-old children with palatal clefts: the impact of additional conditions. *Journal of Communication Disorders*, 66, 1–12. Available from: https://doi.org/10.1016/j.jcomdis.2017.03.001

Flynn, T., Moller, C., Jonsson, R. & Lohmander, A. (2009) The high prevalence of otitis media with effusion in children with cleft lip and palate as compared with children without clefts. *International Journal of Pediatric Otorhinolaryngology*, 73, 1441–1446. Available from: https://doi.org/10.1016/j.ijporl.2009.07.015.

Flynn, T., Lohmander, A., Möller, C. & Magnusson, L. (2013) A longitudinal study of hearing and middle ear status in adolescents with cleft lip and palate. *Laryngoscope*, 123, 1374–1380. Available from: https://doi.org/10.1002/lary.23839

Grant, H.R., Quiney, R.E., Mercer, D.M. & Lodge, S. (1988) Cleft palate and glue ear. *Archives of Disease in Childhood*, 63, 176–179. Available from: https://doi.org/10.1136/adc.63.2.176

Grunwell, P., Brøndsted, K., Henningsson, G., Jansonius, K., Karling, J. & Meijer, M. (2000) A six-centre international study of the outcome of treatment in patients with clefts of the lip and palate: the results of a cross-linguistic investigation of cleft palate speech. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 34(3), 219–229. Available from: https://doi.org/10.1080/02844310050159792

Hall, A., Wills, A.K., Mahmoud, O., Sell, D., Waylen, A., Grewal, S., et al. (2017) Centre-level variation in outcomes and treatment for otitis media with effusion and hearing loss and the association of hearing loss with developmental outcomes at ages 5 and 7 years in children with non-syndromic UCLP: the cleft care UK study. Part 2. *Orthodont Craniofac Res*, 20(suppl 2), 8–18. Available from: https://doi.org/10.1111/ocr.12184

Hammarström, I.L., Nyberg, J., Alaluusua, S., Rautio, J., Neovius, E., Berggren, A., et al. (2020) Scandcleft Project Trial 2: comparison of speech outcome in one and two-stage palatal closure in 5-year-olds with UCLP. *Cleft Palate–Craniofacial Journal*, 57, 458–469. Available from: https://doi.org/10.1177/1055665619888316

Harding, A. & Grunwell, P. (1998) Active versus passive cleft-type speech characteristics. *International Journal of Language & Communication Disorders*, 33, 329–352. Available from: https://doi.org/10.1080/136828298247776

Havstam, C., Dahlgren Sandberg, A. & Lohmander, A. (2011) Communication attitude and speech in 10-year-old children with cleft (lip and) palate: an ICF perspective. *International Journal of Speech Language Pathology*, 13, 156–164. Available from: https://doi.org10.1080/02844310802055102

Hutters, B. & Henningsson, G. (2004) Speech outcome following treatment in cross-linguistic cleft palate studies: methodological implications. *Cleft Palate Craniofacial Journal*, 41(5), 544–549. Available from: https://doi.org/10.1597/02-164.1

John, A., Sell, D., Sweeney, T., Harding-Bell, A., Williams, A. (2006) The cleft audit protocol for speech-augmented: A validated and reliable measure for auditing cleft speech. *Cleft Palate- Craniofacial Journal*, 43(3), 272–288. Available from: https://doi.org/10.1597/04-141.1

Karnell, M. & Van Demark, D. (1986) Longitudinal speech performance in patients with cleft palate: comparisons based on secondary management. *Cleft Palate Journal*, 23, 278–288.

Keuning, K.H.D.M., Wieneke, G.H. & Dejonckere, P.H. (2004). Correlation between the perceptual rating of speech in Dutch patients with velopharyngeal insufficiency and composite measures derived from mean Nasalance scores. *Folia Phoniatrica et Logopaedica*, 56, 157–164. https://doi.org/10.1159/000076937

Klintö, K., Salameh, E.K., Olsson, M., Flynn, T., Svensson, H., Lohmander, A. (2014) Phonology in Swedish-speaking 3-year-olds born with cleft lip and palate and the relationship with consonant production at 18 months. *International Journal of Language & Communication Disorders*, 49(2), 240–54. Available from: https://doi.org/10.1111/1460-6984.12068

Kreiman, J., Gerratt, B.R, Kempster, G.B., Erman, A. & Berke, G.S. (1993) Perceptual evaluation of voice quality: Review, tutorial, and a framework for future reearch. *Journal of Speech and Hearing Research*, 36, 21–40

Kwan, W.M., Abdullah, V.J., Liu, K., van Hasselt, C.A. & Tong, M.C. (2011) Otitis media with effusion and hearing loss in Chinese children with cleft lip and palate. *Cleft Palate–Craniofacial Journal*, 48, 684–689. Available from: https://doi.org/10.1597/10-006.

Liedman-Boshko, J., Lohmander, A., Persson, C., Lith, A. & Elander, A. (2005) Perceptual analysis of speech and the activity in the lateral pharyngeal walls before and after velopharyngeal flap surgery. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 39, 22–32. Available from: https://doi.org/10.1080/02844310510006132

Lohmander, A. (2011) Surgical intervention and speech outcomes in cleft lip and palate. In: Howard S. & Lohmander A. (Eds.). *Cleft Palate Speech: Assessment and Intervention*. UK: Wiley Publ, pp. 55–82.

Lohmander, A., Friede, H., Elander, A., Persson, C. & Lilja, J. (2006) Speech development in patients with unilateral cleft lip and palate treated with different delays of hard palate closure after early velar repair: a longitudinal perspective. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 46, 267–274. Available from: https://doi.org/10.1080/02844310600973621

Lohmander, A., Willadsen, E., Bowden, M., Henningsson, G., Persson, C. & Hutters, B. (2009) Methodology for Speech Assessment in the Scandcleft Project – an international randomised clinical trial on palatal surgery: experiences from a pilot study. *Cleft Palate–Craniofacial Journal*, 46, 347–362. Available from: https://doi.org/10.1597/08-039.1

Lohmander, A., Friede, H. & Lilja, J. (2012) Long-term, longitudinal follow-up of individuals with unilateral cleft lip and palate after the Gothenburg primary early veloplasty and delayed hard palate closure protocol: speech outcome. *Cleft Palate–Craniofacial Journal*, 49(6), 657–671. Available from: https://doi.org/10.1597/11-085

Lohmander, A., Persson, C., Willadsen, E., Lundeborg, I., Alaluusua, S. & Aukner, R., et al. (2017a) Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 4. Speech outcomes in 5-year-olds—velopharyngeal competency and hypernasality. *Journal of Plastic Surgery and Hand Surgery*, 51, 26–36. Available from: https://doi.org/10.1080/2000656X.2016.1254645

Lohmander, A., Hagberg, E., Persson, C., Willadsen, E., Lundeborg, I., Davies, J., et al. (2017b) Validity of auditory perceptual assessment of velopharyngeal function and dysfunction: the VPC-Sum and the VPC-Rate. *Clinical Linguistics & Phonetics*, 31, 589–597. Available from: https://doi.org/10.1080/02699206.2017.1302510

Lohmander, A., Lundeborg, I. & Persson, C. (2017c) SVANTE – the Swedish Articulation and Nasality Test – normative data and a minimum standard set for cross-linguistic comparison. *Clinical*

Linguistics & Phonetics, 31, 137–154. Available from: https://doi.org/10.1080/02699206.2016.1205666

Lohmander, A., & Persson, C. (2008) A longitudinal study of speech production in Swedish children with unilateral cleft lip and palate and two-stage palatal repair. *Cleft Palate Craniofacial Journal*, 45(1), 32–41.

Lohmander, A., Raud Westberg, L., Olsson, S., Tengroth, B. & Flynn, T. (2021) Canonical babbling and early consonant development related to hearing in children with otitis media with effusion with or without cleft palate. *Cleft Palate–Craniofacial Journal*, 58(7), 894–905. Available from: https://doi.org/10.1177/1055665620966198

McWilliams, B.J., Glaser, E.R., Philips, B.J., Lawrence, C., Lavorato, A.S., Beery, Q.C., et al. (1981) A comparative study of four methods of evaluation velopharyngeal adequacy. *Plastic and Reconstructive Surgery*, 68, 1–10. Available from: https://doi.org/10.1097/00006534-198107000-00001

Morrison, M.M., Mason, N.T., Forde, B.L., Stone, P.R., Fowler, P.V. & Thompson, J.M.D. (2022) Speech outcomes of a national cohort of children with orofacial cleft at 5 and 10 years of age. *Cleft Palate–Craniofacial Journal*, 59(11), 1400-1412. Available from: https://doi.org/10.1177/10556656211044939

Nyberg, J., Peterson, P. & Lohmander, A. (2014) Speech outcomes at age 5 and 10 years in unilateral cleft lip and palate after one-stage palatal repair with Minimal Incision Technique—A longitudinal perspective. *International Journal of Pediatric Otorhinolaryng*, 78, 1662–1670. Available from: https://doi.org/10.1016/j.ijporl.2014.07.016

Nyberg, J. & Havstam, C. (2016) Speech in 10-year-olds born with unilateral cleft lip and palate: what do peers say? *Cleft Palate–Craniofacial Journal*, 53, 516–526. Available from: https://doi.org/10.1597/15-140

Pereira, V.J., Tuomainen, J., Lee, K.Y.S., Tong, M.C.F. & Sell, D.A. (2021) A perceptual outcome measure of velopharyngeal function based on the Cleft Audit Protocol for Speech-Augmented (CAPS-A VPC-Sum): validation through a speech osteotomy study. *International Journal of Language & Communication Disorders*, 56, 754–767. Available from: 10.1111/1460-6984.12625

Persson, C., Pedersen, N.H., Hayden, C., Bowden, M., Aukner, R., Vindenes, H., et al. (2020) Scandcleft Project Trial 3: comparison of speech outcome in relation to sequence in two-stage palatal repair procedures in 5-year-olds with UCLP. *Cleft Palate–Craniofacial Journal*, 57, 352–363. Available from: 10.1177/1055665619896637

Peterson, P., Nyberg, J., Persson, C., Mark, H. & Lohmander, A. (2022) Speech outcome and self-reported communicative ability in young adults born with UCLP: comparing results after two methods for palatal repair. *Cleft Palate–Craniofacial Journal*, 59(6), 751–764. Available from: 10.1177/10556656211025926

Peterson-Falzone, S.J. (2013) *Optimal age for palatoplasty to facilitate normal speech development: What is the evidence? Cleft lip and palate*, Springer, pp. 787–802.

R Core Team. (2019) *R: A language and environment for statistical computing*. Vienna, Austria: R Foundation for Statistical Computing. URL Available from: https://www.R-project.org/

Rautio, J., Andersen, M., Bolund, S., Hukki, J., Vindenes, H., Davenport, P., et al. (2017) Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 2. Surgical results. *Journal of Plastic Surgery and Hand Surgery*, 51, 14–20. Available from: https://doi.org/10.1080/2000656X.2016.1254646

Reddy, R.R., Reddy, S.G., Vaidhyanathan, A., Bergé, S.J. & Kuijpers-Jagtman, A.M. (2017) Maxillofacial growth and speech outcome after one-stage or two-stage palatoplasty in unilateral cleft lip and palate. A systematic review. *Journal of Cranio-Maxillo-Facial Surgery*, 45, 995–1003. Available from: https://doi.org/10.1016/j.jcms.2017.03.006

Renfrew, C. (1997) *The Renfrew language scales: Bus story test – a test of narrative speech*. Bicester, Oxon: Winslow Press Limited.

Sand, A., Hagberg, E. & Lohmander, A. (2022) On the benefits of speech–language therapy for individuals born with cleft palate: a systematic review and meta-analysis of individual participant data. *Journal of Speech, Language and Hearing Research*, 65(2), 555–573. Available from: https://doi.org/10.1044/2021_JSLHR-21-00367

Scherer, N.J., D'Antonio, L.L., McGahey, H. (2008) Early Intervention for Speech Impairment in Children with Cleft Palate. *The Cleft Palate Craniofacial Journal*, 45(1), 18–31. Available from: https://doi.org/10.1597/06-085.1

Schönweiler, R., Lisson, J.A., Schönweiler, B., Eckardt, A., Ptok, M., Tränkmann, J., et al. (1999) A retrospective study of hearing, speech and language function in children with clefts following palatoplasty and veloplasty procedures at 18–24 months of age. *International Journal of Pediatric Otorhinolaryngology*, 50, 205–217. Available from: https://doi.org/10.1016/s0165-5876(99)00243-8

Sell, D., Grunwell, P., Mildinhall, S., Murphy, T., Cornish, T.A., Bearn, D., et al. (2001) Cleft lip and palate care in the United Kingdom—The Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. *Cleft Palate–Craniofacial Journal*, 38, 30–37. Available from: https://doi.org/10.1597/15451569_2001_038_0030_clapci_2.0.co_2

Sell, D., Southby, L., Wren, Y., Wills, A.K., Hall, A., Mahmoud, O., et al. (2017) Centre-level variation in speech outcome and interventions, and factors associated with poos speech outcomes in 5-year-old children with non-syndromic unilateral cleft lip and palate: the Cleft Care UK study. *Orthodontics & Craniofacial Research*, 20(Suppl.2), 27–39. Available from: https://doi.org/10.1111/ocr.12186

Semb, G., Enemark, H., Friede, H., Paulin, G., Lilja, J., Rautio, J., et al. (2017) Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 1. Planning and management. *Journal of Plastic Surgery and Hand Surgery*, 51, 1–12. Available from: https://doi.org/10.1080/2000656X.2016.1263

Shaw, W.C., Semb, G., Nelson, P., Brattström, V., Mølsted, K., Prahl-Andersen, B., et al. (2001) The Eurocleft project 1996–2000: overview. *Journal of Cranio-Maxillo-Facial Surgery*, 29, 131–140. Available from: https://doi.org/10.1054/jcms.2001.0217

Shaw, W. & Semb, G. (2017) The Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 11. What next? *Journal of Plastic Surgery and Hand Surgery*, 51(1):88–93. https://doi.org/10.1080/2000656X.2016.1254644

Shaw, W.C., Semb, G. & Lohmander, A. (2015) Clinical outcomes research and evidence-based practice. In: Loose J., Kirschner R. (Eds). *Comprehensive Cleft Care*. USA: Taylor & Francis Group.

Shriberg, L.D. & Kwiatkowsky, J. (1982) Phonological disorders III: A procedure for assessing severity of involvement. *Journal of Speech and Hearing Disorders*, 47, 256–270.

Vallino-Napoli, L.D. (2011) Evaluation and evidence-based practice. In: A. Lohmander & S. Howard (Eds.), *Cleft Palate Speech – Assessment and Intervention* (pp. 317–358). Wiley-Blackwell.

Willadsen, E., Lohmander, A., Persson, C., Lundeborg, I., Alaluusua, S., Aukner, R., et al. (2017) Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 5. Speech outcomes in 5-year-olds—Consonant production and errors. *Journal of Plastic Surgery and Hand Surg*, 51, 37–50. Available from: https://doi.org/10.1080/2000656X.2016.1254647

Willadsen, E., Lohmander, A., Persson, C., Boers, M., Kisling-Moller, M., Havstam, C., et al. (2019) Scandcleft project, Trial 1: comparison of speech outcome in relation to timing of hard palate closure in 5-year-olds with UCLP. *Cleft Palate–Craniofacial Journal*, 56(10), 1276–1286. Available from: 10.1177/1055665619854632

Williams, C., Harding, S. & Wren, Y. (2021) An exploratory study of speech and language therapy intervention for children born with cleft palate + lip. *Cleft Palate–Craniofacial Journal*, 58(4), 455–469. Available from: https://doi.org/10.1177/1055665620954734

Williams, A.C., Sandy, J.R., Thomas, S., Sell, D., Sterne, J.A. (1999) Influence of surgeons's experience on speech outcome in cleft lip and palate. *Lancet*, 13, 1697–1698.

Wren, Y., Miller, L.L., Peters, T.J., Emond, A. & Roulstone, S. (2016) Prevalence and predictors of persistent speech sound disorder at eight year old: findings form a population cohort study. *Journal of Speech Language and Hearing Research*, 59, 647–673. Available from: https://doi.org/10.1044/2015_JSLHR-S-14-0282.

Yang, F.F., McPherson, B. & Shu, H. (2012) Evaluation of an auditory assessment protocol for Chinese infants with nonsyndromic cleft lip and/or palate. *Cleft Palate–Craniofacial Journal*, 49, 566–573. doi:10.1597/09-140.1.

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