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Articulation and velopharyngeal function in patients with cleft lip and/or palate

Outcome predictors

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Abstract

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Cleft lip and/or palate is the most common congenital craniofacial malformation, requiring multidisciplinary treatment, including surgery and often speech therapy. Palatal surgery restores the anatomical barrier between the oral and nasal cavities as well as the palatal function needed for normal speech.

The present thesis aimed to investigate factors thought to impact surgical and speech outcomes. These factors include the timing of surgery, surgical technique, the surgeon's experience, cleft type, and cleft width.

Study I investigated the impact of cleft type and width on velopharyngeal function (VPF) and secondary surgery rates from ages three to 16. Cleft width was associated with increased surgery rates and signs of velopharyngeal insufficiency (VPI) at ages three and five. Contrary to some previous studies, surgery rates and speech outcomes were not associated with cleft type at any age.

Study II examined VPF and articulation development between ages five and ten in children adopted from China compared to Swedish-born children. Adoptees had palatoplasty later and had more severe articulation difficulties at both ages. The number of adoptees and non-adoptees with VPI was similar, but fewer adoptees had competent VPF.

Study III explored different types of intra-velar veloplasty and the impact of cleft width and surgeon experience on outcomes in 5-year-old children. Radical muscle dissection was not superior to intra-velar veloplasty reinforced by the palatopharyngeal muscle. Cleft width had a more significant impact on secondary surgery rates and VPF than did surgical technique—neither affected articulation proficiency.

Study IV examined the association between cleft type and width and surgical and speech outcomes, especially articulation, in 5-year-old children. Cleft width, not type, indicated articulation proficiency. The same errors occurred across all cleft types. Neither cleft type nor width was significantly associated with secondary surgery rates or VPF.

The present thesis highlights the importance of considering various factors when predicting secondary palatal surgery rates and speech outcomes. Cleft width significantly affects secondary surgery rates, VPF, and articulation proficiency, while cleft type and surgical technique do not. Adopted children are at higher risk of persistent articulation errors, which may partly be due to their later palatoplasty.

Keywords: cleft lip and palate, cleft width, cleft type, surgical technique, articulation, velopharyngeal function, international adoption

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Humlan kan inte flyga för vingarna är för små.

Den struntar i allt vad man säger och flyger ändå.

Säg mig du lilla humla hur går den där flykten till?

Men humlan den brumlar och mumlar: Man kan allt man vill.

Lennart Hellsing

To my family

List of Papers

This thesis is based on the following papers, which are referred to in the text by their Roman numerals.

- I. Okhiria AC, Jabbari F, Hakelius MM, Johansson MMB, Nowinski DJ. Greater Palatal Cleft Width Predicts an Increased Risk for Unfavorable Outcomes in Cleft Palate Repair. *Cleft Palate Craniofac J.* 2022;59(8):1030-7.
- II. Okhiria A, Persson C, Johansson MB, Hakelius M, Nowinski D. Longitudinal data on speech outcomes in internationally adopted children compared with non-adopted children with cleft lip and palate. *Int J Lang Commun Disord.* 2023;58(5):1440-53.
- III. Okhiria A, Persson C, Johansson MB, Hakelius M, Jabbari F, Nowinski D. The impact of surgical technique and cleft width on the rate of secondary surgery and velopharyngeal function. (manuscript).
- IV. Okhiria A, Persson C, Hakelius M, Johansson MB, Nowinski D. Associations between cleft type and width and the rate of secondary palatal surgery and articulation proficiency in 5-year-olds with cleft lip and/or palate. (manuscript).

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Abbreviations

BCLP	Bilateral cleft lip and palate
CAPS-A	Cleft Audit Protocol for Speech-Augmented
CAPS-A-AM	Cleft Audit Protocol for Speech-Augmented (American version)
CLP	Cleft lip and/or palate
CPO	Cleft palate only
CSCs	Cleft speech characteristics
DSCs	Developmental speech characteristics
IA	Internationally adopted
NA	Non-adopted
PCC	Percent of correct consonants
PCC-A	Percent of correct consonants – adjusted for age
RS	Robin Sequence
SCP	Cleft in the soft palate
SHCP	Cleft in the soft and hard palate
SLP	Speech and language pathologist
SVANTE	The Swedish Articulation and Nasality Test
UCLP	Unilateral cleft lip and palate
VPC	Velopharyngeal competence
VPI	Velopharyngeal insufficiency
VPF	Velopharyngeal function

Introduction

Cleft lip and/or palate (CLP) is the most common congenital craniofacial deformity of the face that arises during the fourth to eleventh weeks of pregnancy, with an incidence of 1/700 births with ethnic and geographic variation (1). In Sweden, the incidence has been reported to be 2/1000 births, meaning that 150-200 children with CLP are born every year (2). The cleft can be uni- or bilateral and comprise only the lip with or without alveolus or lip, alveolus, and palate. A cleft palate can also occur without affecting the lip and alveolus. The extent of the palatal cleft varies from a submucous, occult cleft with split uvula to an overt cleft, including the entire soft and hard palate up to the incisive foramen (3). For clefts involving the palate; see Figure 1.

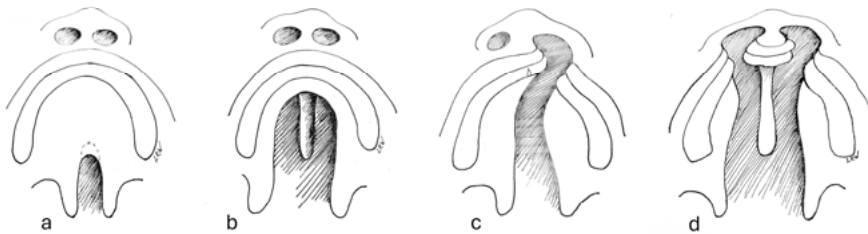


Figure 1. a) cleft affecting the soft palate (SCP), b) cleft affecting the soft and hard palate (SHCP), c) unilateral cleft lip and palate (UCLP), and d) bilateral cleft lip and palate (BCLP). *Illustrations by Liisi Raud Westberg*

Cleft lip and/or palate affects a child's eating ability during infancy, speech development, ear function and hearing, facial growth, dental occlusion, psychosocial well-being, and facial appearance. Because of the many different symptoms, multidisciplinary care is necessary to treat this condition. In Sweden, children with clefts are treated from birth to at least 19 years of age by an interdisciplinary team consisting of, among others, plastic surgeons, nurses, orthodontists, maxillofacial surgeons, speech and language pathologists (SLPs), psychologists, and phoniatricians. All children in Sweden are treated at one of the six academic medical centers. In many respects, the care is similar regardless of which center the child is treated at. However, the surgical treatment of the palate differs; the palate is closed either in a 1-stage procedure,

with both soft and hard palate closure at nine to 15 months, or in two stages, with soft palate closure at six months and hard palate closure at two years of age.

Routine follow-ups have been harmonized nationally at 18 months, three, five, ten, 16, and 19 years. At routine follow-ups, a speech assessment is conducted by an SLP specialized in cleft palate speech, following a standardized procedure. Standardized speech material, the Swedish Articulatory and Nasality Test (SVANTE) (4), is used at all ages except for follow-up at 18 months. When possible, spontaneous speech is elicited. All speech is recorded, and articulation and velopharyngeal function (VPF) are analyzed. To enable comparisons and ensure equal care across centers, the orthodontists' and SLPs' assessments are registered in the Swedish cleft lip and palate registry as part of routine follow-ups. Surgical data are recorded continuously.

Background

Surgical treatment of CLP

Surgical treatment of the cleft varies both nationally and internationally. Despite extensive research, conclusive evidence regarding the method and best timing for soft and/or hard palate closure is lacking. Most centers worldwide close the cleft palate in one session, while four out of six CLP centers in Sweden apply a 2-stage procedure. Surgery in two stages was developed to balance speech outcomes and maxillary growth. Early closure of the palate, especially the soft palate, is generally considered to favor speech development and VPF (5-8). However, for maxillary growth, later closure of the hard palate seems to be preferable (9-12).

A review showed a need for more comparative studies of different surgical methods to investigate the connection between speech outcomes and different surgical procedures (13). The Scandcleft trials compared four different surgical protocols, with 1- or 2-stage palatoplasty and different timing for the soft and hard palate closure. The rate of secondary palatal surgery, facial growth, and speech outcomes up to ten years of age were analyzed (14-20). Although there were differences in some respects between the protocols, none appeared to be decisively superior to the others. Regarding the speech results from the Scandcleft trials, no significant differences were observed at age five, except for the occurrence of retracted articulation (e.g., /t/ is articulated as /k/). This error was more frequently observed in children who underwent hard palate closure at three years than those with hard palate closure at one year. Both groups had soft palate closure at three to four months (16). At ten years of age, patients with hard palate closure at three years of age had the lowest percentage of correct consonants (PCC). However, this group also had the highest rate of primary velopharyngeal competence (a competent velopharyngeal function without having had speech-improving surgery) (20).

Apart from timing (21-24) and whether 1- or 2-stage surgery is performed, details of the surgical technique may also be of importance to outcomes (25, 26). Radical muscle dissection of the soft palate, as described by Sommerlad (27, 28), has been shown to reduce the secondary surgery rate and improve velopharyngeal function and articulation (29-31). Also of importance is the surgeon's experience (32). Using Sommerlad's technique, the rates of secondary velopharyngeal surgery decreased from 10.2% to 4.6% in a group of patients operated on consecutively by the same surgeon during a 15-year period

between 1978 and 1992 (27). Subsequently, in the following cohort operated on by the same surgeon between 1993 and 2006, there was a decrease in fistula repairs, but the rate of speech-improving surgery remained stable (33).

At Uppsala University Hospital, primary lip surgery is performed at age three to four months, as described by Skoog (34) for unilateral clefts. Bilateral cleft lip repair is performed at age three to six months, as described by Mulliken (35). A 2-stage procedure for closing the palate has been used since the mid-1970s, and from the mid-1984, soft palate closure has been performed at six months and hard palate closure at two years.

Before 2007, the soft palate was closed with an intra-velar veloplasty reinforced by the palatopharyngeal muscle (36). In 2007, more extensive levator muscle dissection was introduced and progressively developed towards a radical muscle dissection. In August 2009, radical muscle dissection, originally described by Sommerlad (27, 28), was introduced and has been used exclusively ever since. Soft palate closure was initially performed under 3.5 times loop magnification but has since 2014 been performed under a surgical microscope with 2.3-14 times magnification. Usually, 6-10 times magnification is used. The soft palate closure is performed through incisions at the borders of the cleft without lateral releasing incisions. In most cases, an anterior-based vomerian flap is used to reinforce the nasal layer of the soft palate.

The residual cleft in the hard palate is repaired in two layers. If needed for closure without tension, lateral gingival releasing incisions along the cervical lines of the teeth are performed, as described by von Langenbeck (37).

The alveolar cleft is reconstructed with secondary bone grafting from the iliac crest at the mixed dentition stage in conjunction with orthodontic treatment (38, 39).

Regardless of how and when surgery is performed, secondary palatal surgery may be required due to a palatal fistula or velopharyngeal insufficiency (VPI). If the speech assessment indicates VPI and if VPI is confirmed by nasoendoscopy, a speech-improving surgery may be indicated. The most common speech-improving surgery is the superiorly based pharyngeal flap, but in some cases, a re-repair of the soft palate is performed using the Sommerlad technique. In rare cases, speech-improving surgery with buccinator flaps is performed. The choice of method for speech-improving surgery is made in consultation with a phoniatrician and depends on the configuration and muscular activity of the soft palate and pharyngeal walls, as visualized by nasoendoscopy.

Speech in children with CLP

Speech production is a complex process involving several structures, including the vocal cords, soft palate, tongue, and lips. The ability to achieve velopharyngeal closure, i.e., separating the oral and nasal cavities, is a

prerequisite for the interaction between phonation, resonance, and articulation to function adequately (40, 41). The production of consonants can be voiced or voiceless, with the airstream being obstructed mainly in the oral cavity using the lips and tongue (42). Due to structural deviances, the development of speech and speech sounds can be affected by CLP, and one of the primary goals of cleft palate surgery is to facilitate normal speech. The onset of babbling may be delayed and children with CLP produce fewer high-pressure consonants than children without a cleft palate do (7, 43-45). In children with unoperated clefts, babbling is dominated by nasals, approximants, and glottal sounds (43, 45). Speech difficulties may remain even after palatal closure, requiring speech therapy and/or speech-improving surgery. In children with cleft palate, articulation is often impaired, and studies have shown that more than 50% of children with CLP require speech therapy (46). The velopharyngeal function (VPF) may also be impaired, and the reported rate of speech-improving surgery is between 2.6%-33.3% (33, 47-49). There are various reasons why the figures differ from study to study. It can be due to methodological differences, the surgical technique and skill, the team's threshold to perform speech-improving surgery, the preferences of patients and their families, and the age at the time of follow-up.

Speech errors related to the cleft palate can be divided into passive and active speech errors (50). Passive speech errors include hypernasality, audible nasal air leakage, and weak pressure consonants related to an oro-nasal coupling, i.e., fistula, residual cleft in the alveolus, or VPI, all of which require surgery. Active speech errors, often referred to as cleft speech characteristics (CSCs), can be further subdivided into oral and non-oral articulation (51). Oral retracted articulation is a common articulation error in children with CLP (52), especially when delayed hard palate closure in 2-stage surgery is practiced. It is thought to be a compensatory strategy to avoid nasal air leakage through an oro-nasal coupling, such as a residual cleft in the alveolus or hard palate, and the articulation is moved to a place behind the air leakage where adequate intra-oral pressure can be obtained (Figure 2b). This means that speech sounds produced anteriorly, for example, /t/, may be produced further back in the mouth, i.e., retracted to a palatal, velar, or uvular place of articulation where it is possible to obtain enough intra-oral pressure for plosives.

Non-oral articulation often occurs due to inadequate ability to produce intra-oral pressure (50, 53) and, thus, inability to produce high-pressure consonants, i.e., plosives such as /k/ or fricatives such as /s/. The child tries to compensate for the inability to produce high-pressure plosives and fricatives by retracting the articulation and making the consonant at the vocal cord level, so-called glottal articulation (Figure 2c), or producing a fricative through the nose, so-called active nasal fricative. These errors are often a consequence of VPI, which requires speech-improving surgery. However, the production of /s/ as a nasal fricative does not always indicate VPI but may instead be a case of mislearning. Non-oral as well as oral errors can remain after successful

surgery, as it is often a well-established articulation manner. Consequently, speech therapy may also be necessary. Correct and incorrect places of articulation are shown in Figure 2.

Other articulation errors often noted in children with CLP are so-called developmental speech characteristics (DSCs), which are classified according to phonological processes. It is worth noting that those speech errors, such as stopping of fricatives (e.g., where /s/ is replaced with /t/) or fronting (e.g., where /k/ is replaced by /t/), are not only common in children with CLP but also in typically developed children without CLP at certain ages. This indicates that such errors are not usually related to any structural anomaly but rather to an immature and developing sound system.



Figure 2. a) correct place of dental/alveolar articulation, b) dental articulation retracted to velar articulation place, and c) articulation retracted to vocal cord level (glottal articulation). *Illustrations by Liisi Raud Westberg*

Speech assessment

It is essential to use standardized speech material developed to capture the speech characteristics of interest when assessing speech, both for clinical and for research purposes. SVANTE (4) is an example of a test designed to enable a detailed speech assessment in children with CLP and has been used nationally in Sweden since 2005. It consists of 74 words and 13 sentences. Fifty-nine of the words include target consonants particularly vulnerable to cleft-related speech difficulties (plosives /p/, /b/, /t/, /d/, /k/, /g/, and voiceless fricatives /f/, and /s/ in initial, medial, and final position, and the voiceless fricative /ç/ in initial position). The first nine words also include high vowels (/i:/, /u:/, and /ɤ:/) for the assessment of hypernasality. Five words include nasals /m/ and /n/ for assessment of hyponasality, and the final ten words include s-clusters. The 13 sentences consist of eight sentences with high-pressure consonants, two with low-pressure consonants, two with oral and nasal consonants, and one with nasal consonants.

Other examples of standardized speech materials are the Cleft Audit Protocol for Speech-Augmented (CAPS-A) developed in the United Kingdom and Ireland (54) and the American version of CAPS-A, CAPS-A-AM (55). There is a critical difference between SVANTE and CAPS-A and CAPS-A-AM in how they categorize consonant errors. In SVANTE, they are split into two categories - oral and non-oral (4). However, in CAPS-A and CAPS-A-AM, errors are divided into three categories - anterior oral, posterior oral, and non-oral. (54, 55). In addition, the scales and the scale definitions differ. The differences between standardized speech materials must be considered when comparing and interpreting results across studies.

To avoid biases and to ensure reliability, the gold standard for assessments in research involves blinded re-assessment by at least two independent specialist SLPs using high-quality audio recordings (56, 57). To ensure equal assessments, re-assessments should be preceded by calibration (51, 55, 57, 58).

Speech outcome measurements

Depending on the research question, various outcome measurements can be utilized. The present thesis focused on articulation, which can be assessed at varying levels of precision, and velopharyngeal function. In Study II-IV in the present thesis, we used percent of correct consonants (PCC) to measure consonant proficiency. PCC was initially developed by Shriberg and Kwiatkowski (59) to measure the percentage of correct consonants in conversational speech. A modified version in single words has since been developed and used to assess articulation proficiency in children with CLP (60-62). When using PCC, all articulation errors are weighed equally, regardless of age appropriateness. To account for this, PCC adjusted for age (PCC-A) (63) can be calculated. In PCC-A, age-appropriate s-distortions such as inter-dental, lateral, supra-dental, retroflex, alveolo-palatal, and palatal production of /s/ are scored as correct. In both PCC and PCC-A, signs of VPI are disregarded.

As PCC or PCC-A does not reveal the type of articulation errors made, an error analysis can be performed. A common way to do this is to divide errors into different CSCs. How errors are categorized differs; for instance, retracted articulation of anterior consonants to a palatal place is differentiated from retracted articulation to a velar or uvular place in, for example, Baillie and Sell and in Butterworth et al. (33, 64) but not in, for example, the Scandcleft trials (16, 17). One reason for not differentiating between palatal and velar articulation is that it may be hard to distinguish them, causing agreement to decrease. The categorization of CSCs applied in the present thesis and the studies included are the same as those in the Scandcleft trials (16, 17); see Table 1.

Table 1. Categorization of active cleft speech characteristics (CSCs).

Oral errors	Non-oral errors
Retracted/backed to palatal/velar/uvular place of articulation	Glottal stop and fricative
Double articulation, e.g., articulate /p/ and /k/ simultaneously	Pharyngeal stop and fricative
	Nasal for unvoiced stop or fricative
	Active nasal fricative

Developmental speech characteristics (DSCs) have been used to also capture any development-related errors in children with CLP (16, 17, 62, 65). Such errors include, for example, frication (a plosive replaced with a fricative), velar fronting (a velar consonant is replaced with a dental or alveolo-dental consonant), stopping (a fricative is replaced with a plosive), and voicing errors (difficulties differentiating between voiced and voiceless consonants) and are considered DSCs. Using the same definition of DSCs as in the Scandleft trials, the decision was made to use DSCs in the present thesis as well. An articulation error can be classified as both a CSC and DSC if, for example, /s/ is produced as /k/, which involves stopping and retracted articulation simultaneously.

Velopharyngeal function (VPF) can be assessed from perceptual ratings of hypernasality, audible nasal emission, and weak pressure consonants or as an overall rating of VPF. In Study I, variables associated with VPF were used: ratings of hypernasality and audible nasal emission and the presence or absence of glottal articulation. In Study II-IV, two different measurements for assessing VPF were used: rating of perceived velopharyngeal competence (VPC) in sentences (VPC-R) (Study II and III) and a composite score for velopharyngeal competence (VPC-Sum) (Study II-IV) based on single words (66).

Influencing factors

As described in the surgical treatment section, various factors are believed to affect different outcome measurements. The age at surgery, surgical technique, surgeon's experience, and whether the surgery is performed in one or two stages are some care-related factors we can strive to develop and control for. However, there are several factors we cannot control for, including patient-related factors such as cleft type, cleft width, and cleft extent into the hard palate in those with cleft palate only (CPO). All are suggested to affect outcomes but are only partially clarified.

Studies have shown that the more extensive the cleft, the more articulation difficulties. That is, children with cleft in the soft palate (SCP) have the best

articulation, followed by those with cleft in the soft and hard palate (SHCP), then unilateral cleft lip and palate (UCLP), and lastly, bilateral cleft lip and palate (BCLP) (33, 64, 67-69). However, regarding VPF, it does not seem to follow the same pattern, and the literature contains inconsistencies regarding VPF and the rate of speech-improving surgery. The rate of speech-improving surgery has been shown to follow the severity of the cleft in some studies (70, 71), but others have reported an inverted relation with higher incidences in individuals with CPO (72, 73). Studies have shown that children with SCP experience fewer issues with VPF than those with other cleft types. However, the cleft type that had most issues varied across the studies (67, 69, 74). A meta-analysis of 383 studies showed that when all studies reporting normal resonance outcomes were combined, individuals with SCP had the best results, followed by UCLP, SHCP, and BCLP (75). When the severity of CPO was investigated, it was shown that children with SCP had better VPF than children with SHCP (64, 76-78).

The impact of cleft width has rarely been investigated in relation to speech outcomes but more commonly in relation to surgical outcomes such as fistula rate or the rate of speech-improving surgery. Increased cleft width has been related to increased rates of surgery due to fistulas and VPI (74, 79-81). The one study relating cleft width to assessments of VPF found a weak but significant association between VPC-R and posterior cleft dimensions at the level of tuberosity (82).

The rationale for the present studies

Achieving normalized speech is a crucial objective of cleft palate surgery. Nevertheless, there are varying practices regarding the optimal timing and approach if palatal surgery is to achieve the most favorable outcome. Studies comparing various surgical techniques have frequently been hampered by the surgery being conducted at different ages or that the assessments afterwards are performed at different ages, making it challenging to compare the techniques accurately. Hence, improved studies are needed to compare different techniques where the surgery across techniques is performed at the same age with the same 1- or 2-stage procedure protocol and the same age at follow-up.

Longitudinal studies are still scarce as most studies are conducted during preschool age. However, articulation continues to develop beyond this age, and velopharyngeal function may deteriorate over time. Moreover, individuals with CLP undergo continuous treatment during childhood and adolescence, which can affect their speech. Therefore, more longitudinal studies are needed to track speech development over time and to better understand the effects of treatment on speech outcomes.

Besides the timing and method of the surgery, cleft width and cleft type have been proposed to affect the rate of secondary palatal surgery, velopharyngeal function, and articulation. Rarely have articulation and velopharyngeal function been used as outcome measures in investigations of cleft width. Instead, surgical outcomes such as fistula rate or the rate of speech-improving surgery have been in focus. While these outcome measurements are important, they do not measure actual speech outcomes. Consequently, they do not provide a complete understanding of how cleft width impacts articulation and velopharyngeal function, which may be impaired but not to such an extent that speech-improving surgery is required. Although including all cleft types in studies has become more common, we still need to fully understand the impact of cleft type on speech. Additionally, the combined effect of cleft width and type on speech outcomes is yet to be investigated.

Aims

Overall aim

The present thesis focuses on investigating how patient-related (cleft width and type) and care-related (surgical technique, surgeon experience, adopted vs non-adopted) factors impact speech and surgical outcomes.

Study I

The first study aimed to investigate the impact of cleft width and cleft type on the rate of secondary surgery and speech variables related to velopharyngeal function from a longitudinal perspective.

Study II

The primary aim of this study was to investigate how velopharyngeal function and articulatory ability develop longitudinally between ages five and ten in children with CLP adopted from China compared to a matched cohort of children with CLP born in Sweden.

Study III

This study aimed to assess the efficacy of different types of intra-velar veloplasty in an otherwise uniform surgical protocol and to evaluate the influence of cleft width and the surgeon's experience on outcome measurements.

Study IV

This study aimed to investigate the association between cleft type and width and secondary palatal surgery, articulation proficiency, and velopharyngeal function, as well as to determine whether cleft type influenced the type of articulation errors.

Ethical approval

I - IV. Ethical approval was obtained from the Regional Ethical Review Board, Uppsala University, Dnr: 2017/457.

Methods

Participants

Study I

Patients treated for cleft lip and/or palate at Uppsala University Hospital, born from 1984 to 2002, treated with the same surgical technique in a 2-stage palatal surgery, and followed by the CLP team from birth up to 16 years of age were reviewed. Out of a consecutive series of 313 patients, 213 individuals were included: 86 with UCLP (58 boys, 28 girls), 37 with BCLP (31 boys, six girls), and 90 with SHCP (38 boys, 52 girls). Nine included patients were diagnosed with a syndrome (four with BCLP, three with UCLP, and two with SHCP had syndromic Robin sequence (RS)). Thirteen children with SHCP had non-syndromic RS.

One hundred patients were excluded due to a) missing data, i.e., missing medical records or dental cast (n=27); b) transfer of care to another cleft center (n=12); c) not treated according to the original surgical protocol (n=9); d) operated on using 1-stage palatoplasty (n=11); e) cleft in the soft palate only, i.e. only requiring one primary operation (n=20); f) cleft in lip and palate but not in the primary palate (n=4); g) inadequate speech for reliable assessment of VPC (n=7); h) death (n=10). Of those, four were diagnosed with a syndrome, two with syndromic RS, and eight had non-syndromic RS; see Figure 3.

Study II

Internationally adopted children (IA) from China born from 2000 to 2009 and treated for UCLP or BCLP at Uppsala University Hospital were reviewed. A total of 39 individuals met the inclusion criteria. Among them, 16 were excluded due to incomplete audio recordings. Twenty-three children were included in the study, 17 with UCLP and six with BCLP, six girls and 17 boys. The IA children were matched with non-adopted, Swedish-born children (NA) according to cleft type, gender, and surgical technique for the soft palate. If more than one NA child with a complete audio recording matched an IA child, a random choice was made to prohibit bias selection.

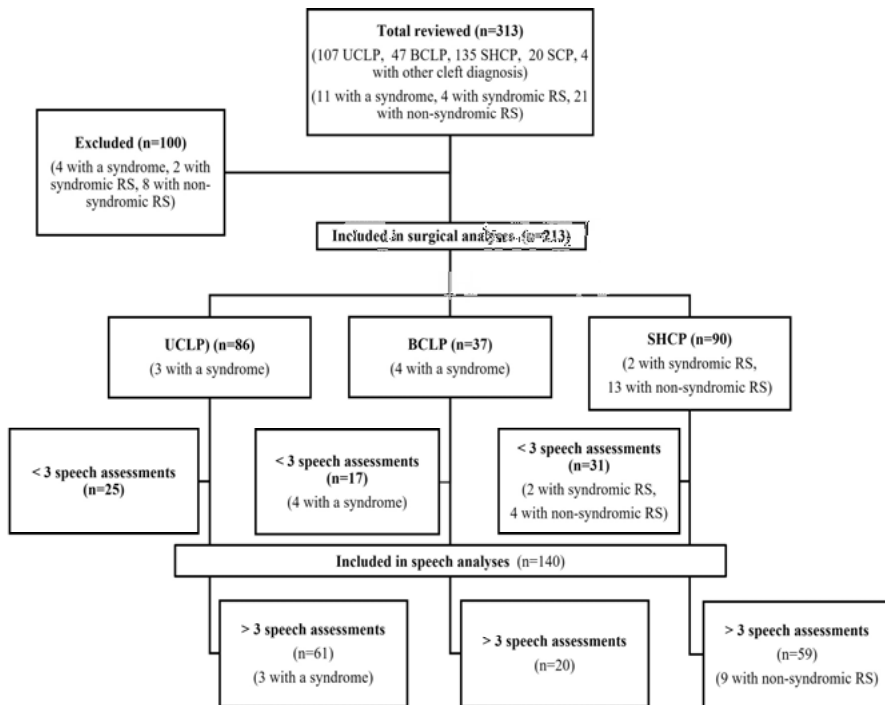


Figure 3. Flowchart of included and excluded individuals in Study I. UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, SCP = cleft in the soft palate, RS = Robin sequence.

Study III and IV

A consecutive series of 356 children born 2000-2015 with cleft lip and/or soft and hard palate treated at our department from birth were reviewed at five years of age. One-hundred and fifty-five met the inclusion criteria of being non-syndromic and having had 2-stage palatoplasty with soft palate closure at six months (-2, +3 months) and hard palate closure at 24 months (+/-3 months). Fifty-five children were excluded due to 1) missing or incomplete data (n=40) or 2) having an additional diagnosis that may affect speech and/or language development (n=15). This resulted in 100 children: 62 UCLP (19 girls and 43 boys), 17 with BCLP (six girls and 11 boys), and 21 individuals with SHCP (15 girls and six boys). The individuals with UCLP were included in both Study III and IV, while individuals with BCLP and SHCP only were included in Study IV.

A total of 201 individuals did not meet the initial inclusion criteria. Among them, 111 did not meet the criteria of 2-stage surgery or the age at surgery. Eighty-six of these individuals had SHCP, of which 74 were treated with 1-stage palatoplasty. Other reasons for exclusion were having moved to another region or being deceased; see Figure 4.

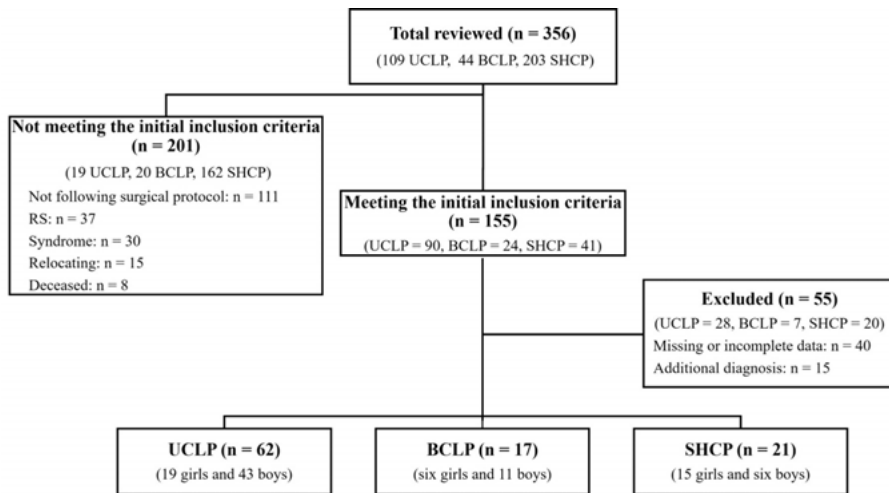


Figure 4. Flowchart of included and excluded individuals in Study III and IV. UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, RS = Robin sequence.

Surgical treatment

Study I

Since 1984 and onwards, the surgical protocol has included a 2-stage palatal surgery. The soft palate closure between 1984 and 2002 was performed at six months of age with an intra-velar veloplasty reinforced by the palatopharyngeal muscle (36). By releasing the muscle and nasal layer from the posterior edge of the hard palate without separating the levator muscle from the nasal mucosa, intra-velar muscle retro-positioning was achieved.

At two years of age, the hard palate was closed in two layers. As described by von Langenbeck (37), relaxing incisions inside the dentition were used if needed. A pharyngeal flap or re-repair of the soft palate was used for speech-improving surgery.

Study II-IV

Different surgical techniques for closure of the soft palate have been used during the study period. Prior to 2007, the soft palate was closed using intra-velar veloplasty reinforced by the palatopharyngeal muscle (36) (in Study III, referred to as “non-dissection”). More extensive levator veli palatini muscle dissection was introduced in 2007 and gradually evolved towards more radical muscle preparation and retro-positioning (in Study III, referred to as “transition”). Since August 2009, radical muscle dissection, as described by Sommerlad (27, 28), has been performed consistently (in Study III, referred to as “Sommerlad”). Four surgeons were involved during the study period. The two

senior surgeons (Surgeon 1 and 2) performed the soft palate closure without loop magnification, while the two more junior surgeons (Surgeon 3 and 4) performed the surgery under 3.5 loop magnification. Since 2014, all soft palate closures were carried out by Surgeon 3 and 4 under a microscope with 2.3-14 times magnification. Usually, 6-10 times magnification is used.

The hard palate was closed in two layers at the age of two years using releasing incisions if necessary to avoid tension, as described by von Langenbeck. (37). When using releasing incisions, the incision lines were always placed along the cervical lines of the teeth.

Depending on the findings from the visualizing assessment, speech-improving surgery was performed with re-repair of the soft palate or a pharyngeal flap. The patients were evaluated using speech assessment, nasoendoscopy, and, when possible, nasometry before speech-improving surgery.

Measurement of cleft width

Study I, III, and IV

For Study I, III, and IV, cleft width was measured on dental study casts obtained at the time of the first surgical procedure, either the lip-plasty for those with UCLP and BCLP or the soft palate closure for those with SHCP. Descriptions of the reference points and linear measurements have previously been published (83-85). T-T1 represents the posterior width of the alveolar arch in the tuber area, and A-A1 represents the width of the cleft at the level of T-T1. Cleft width was calculated as the ratio $A-A1/T-T1$ (Figure 5), and distances were measured to the nearest 0.01 mm using a digital caliper by an orthodontist with extensive experience. Dental casts were randomly chosen for repeated measurements to establish intra-rater reliability, 15% in Study I and 30% in Study III and IV.

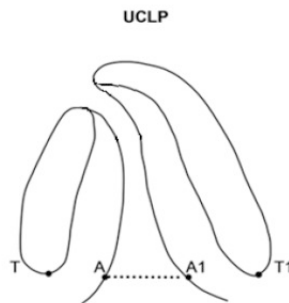


Figure 5. Schematic drawing of an infant maxillary dental cast with unilateral cleft lip and palate (UCLP). T-T1 = maxillary arch width, measured as the transverse distance between the tuberosity points on each side. A-A1 = width of the cleft at the level of T-T1.

Speech material and assessment

Study I

Documented speech assessments of hypernasality, audible nasal emission, and glottal articulation at three, five, ten, and 16 years were retrieved from the medical record. The assessments were analyzed and used in statistical analyses. A total of 492 assessments were made for the 140 individuals with three or four assessments. To assess reliability, data from the medical records were compared with re-assessments of audio recordings from ten percent (n= 49) in each cleft group. Thirty percent of the randomly chosen audio recordings were duplicated to calculate intra-rater agreement. One independent SLP conducted the re-assessments.

Until 2016, hypernasality and audible nasal emission were rated on a 5-point ordinal scale, which then changed to a 4-point ordinal scale. Results will be presented on a 4-point scale (0 = normal, 1 = mild, 2 = moderate, and 3 = severe). To allow for this, scale steps 0 and 1 from the 5-point scale were pooled into scale step 0 on the 4-point scale. Glottal articulation was also rated on the ordinal scale until 2000, but subsequently, it has been transcribed and reported in number of occurrences. For this study, glottal articulation was dichotomized into absent or present. It was considered absent if rated with scale steps 0-1 on the 5-point scale or transcribed with a maximum of two occurrences.

Study II-IV

Recording and editing

Words and sentences from SVANTE (4) were recorded at ages five (Study II-IV) and ten (Study II). The words were elicited through picture naming or reading from a list, and the sentences were repeated after the SLP. The audio recordings were made with Zoom H4n or a PC with Soundswell software (Saven Hitech, Stockholm, Sweden) and a condenser microphone (Røde NT4, Sydney, Australia or Philips SpeechMike Classic 6264Details).

Using Praat software (86), three sets of speech material were edited from the recordings: 1) Fifty-nine single words, each including one target high-pressure consonant (plosives /p/, /b/, /t/, /d/, /k/, and voiceless fricatives /f/, and /s/ in initial, medial and final position, and the voiceless fricative /θ/ in initial position); 2) A nine-word string (the first nine monosyllabic words edited to a string of words with no pause between them) including high vowels (/i:/, /u:/ and /æ:/) for assessment of hypernasality; 3) Twelve sentences for perceptual assessment (ten including oral consonants, two including oral and nasal consonants) of velopharyngeal competence (VPC-R). The first eight sentences each included four occurrences of high-pressure target consonants and were also used for transcription in Study II. All edited audio files were randomized and assigned codes to enable blinded assessments.

Calibration

Shortly before conducting assessments in Study II-IV, calibration sessions were held to ensure equal assessments. Audio recordings not included in the studies were used.

Prior to Study II, the two transcribers spent four hours transcribing 14 audio recordings individually and compared and discussed their transcriptions immediately after each audio recording. In a second session, the three raters of hypernasality and VPC-R spent one and a half hours rating 14 audio recordings, one at a time, and then discussed their individual ratings. In cases of disagreement, they listened to the audio recording again and discussed until reaching consensus.

Thirteen audio recordings were rated for hypernasality and VPC-R, and four were transcribed prior to the calibration sessions of Studies III and IV. In the first calibration session of approximately one and a half hours, the three raters compared their ratings, and in cases of disagreement, the audio recording was listened to again, and the raters discussed until reaching consensus. The two SLPs also doing the transcriptions held a second 90-minute calibration session to review and discuss their transcriptions. In cases of disagreement, they listened to the audio recording again followed by a discussion and reaching consensus.

Assessment

All assessments were carried out in a blinded manner to avoid any biases. The SLPs rated and transcribed the audio recordings individually, using high-quality headphones. They were able to listen to each audio recording as many times as needed. It is worth noting that the transcribers in Study II differed from those in Study III and IV. Similarly, the independent SLPs who rated hypernasality and VPC-R in Study II also differed from those in Study III and IV.

As individuals with UCLP in Study III were also included in Study IV, the speech assessments were conducted simultaneously for all 100 participants to ensure equivalent assessments, forming the basis for Study III and IV, as were cleft width measurements.

Phonetic transcriptions

Words and sentences from SVANTE (4) were transcribed from audio recordings made at routine follow-ups at ages five and ten. Semi-narrow transcriptions – defined as a phonetic transcription using the International Phonetic Alphabet (IPA) (87) and the symbols for nasal escape, velopharyngeal friction, weak articulation, voicing, devoicing, and active nasal fricative from the extended IPA symbols for disordered speech (88) – were performed. Consonant phonemes investigated in the present thesis were bilabial, dental, and velar plosives, and the fricatives /s/, /f/, and /θ/.

In Study II, one local SLP transcribed the whole material, and an independent SLP transcribed 50% from each age to calculate inter-transcriber agreement. Thirty percent of the audio recordings in each group at each age were randomly selected and re-assessed to calculate intra-transcriber agreement.

In Study III and IV, the whole material was transcribed by one independent SLP and the author. The transcriptions made by the independent SLP were used as the results for all participants. Thirty percent were randomly selected and re-transcribed to calculate intra-transcriber agreement.

Assessment of velopharyngeal function and hypernasality

Velopharyngeal function (VPF) was assessed using one or two variables: rating of perceived velopharyngeal competence in sentences (VPC-R) (66) and a composite score for velopharyngeal competence (VPC-Sum) for single words (14, 66). VPC-R is rated on a 3-point ordinal scale (0 = competent, 1 = marginally incompetent, and 2 = incompetent). VPC-Sum is a composite score including a) perceptual ratings of hypernasality rated on a 4-point ordinal scale (0 = normal, 1 = mild, 2 = moderate, 3 = severe), b) perceptual signs of VPI from transcriptions (nasal emission and weak pressure consonants), and c) active non-oral speech errors from transcriptions, used to assess VPF at the word level. The three variables generate a score of 0-2, which is then summed to calculate VPC-Sum; see Table 2. The interpretation of VPC-Sum was used in the statistical analyses.

Hypernasality and VPC-R were rated by three SLPs, two independent and the author. The score for each child was classified based on the majority decision of the three SLP ratings. In case of total disagreement, the middle value was chosen. Thirty percent in Study II-IV were re-assessed to calculate intra-rater agreement.

Table 2. Transfer rules for values from the assessment of hypernasality, active non-oral errors, and symptoms of velopharyngeal insufficiency (VPI) (audible nasal emission, weak pressure consonants, and nasal realization of voiced consonants) into summary scores of the VPC-Sum.

Variables	Transfer rules			Summary
Hypernasality (ordinal 4-point scale)	0 = 0	1 = 1	2-3 = 2	0-2
Active non-oral errors (number from phonetic transcription)	0-2 = 0	3-5 = 1	≥6 = 2	0-2
VPI symptoms (number from phonetic transcription)	0-2 = 0	3-5 = 1	≥6 = 2	0-2
VPC-Sum*				0-6

*Interpretation VPC-Sum. 0-1 = 0: Competent. 2-3 = 1: Marginally incompetent: evidence of minor problems suggesting borderline closure. 4-6 = 2: Incompetent: evidence of significant problems usually requiring surgical management. VPC = velopharyngeal competence.

Speech outcomes

The percent of correct consonants (PCC) was calculated based on the transcriptions. In Study II, PCC was calculated for both the word and sentence levels. In addition, PCC adjusted for age (PCC-A) (63) was calculated in Study IV. When using PCC-A, s-distortions such as inter-dental, lateral, supra-dental, retroflex, alveolo-palatal, and palatal production of /s/ are regarded as age-appropriate at fine years of age and scored as correct. In Study II and IV, articulation errors were divided into CSCs and DSCs, as described in Willadsen et al. (16, 17). CSCs were further divided into oral and non-oral errors. Three or more occurrences signified an error. In Study II, s-errors (inter-dental, lateral, supra-dental, retroflex, alveolo-palatal, and palatal production of /s/) were reported separately, although they may be categorized as CSCs or DSCs because the main transcriber only used the sign for alveolo-palatal fricative (ɛ) for both alveolo-palatal (ɛ) and palatal fricatives (ç). In Study IV, s-errors were divided into CSCs and DSCs. VPC-R was used in Study II and III, while VPC-Sum was used in all three studies. The speech variables used in Study II-IV are presented in Table 3

Table 3. Speech variables used in Study II-IV.

Outcome	Study II	Study III	Study IV
PCC	X	X	X
PCC-A			X
CSCs	X		X
DSCs	X		X
VPC-R	X	X	
VPC-Sum	X	X	X

PCC = percent of correct consonants, PCC-A = PCC – adjusted for age, CSCs = speech cleft characteristics, DSCs = developmental speech characteristics, VPC = velopharyngeal competence.

Statistical analyses

All statistical analyses were conducted in collaboration with professional statisticians. Logistic regression analyses were performed in Study I, III, and IV to investigate the impact of cleft width, cleft type, and surgical technique on the rate of secondary palatal surgery. Linear regression analysis estimated the association between cleft type and width with PCC and PCC-A in Study IV. Ordinal regression analysis was used in Study III and IV to investigate the association between surgical technique, cleft type and width, and VPF. In Study III, Spearman's rank order correlation test calculated the correlation between cleft width and PCC. Differences between groups were assessed using non-parametric analyses. The results on differences between surgical techniques for the surgeon active throughout the study period will be presented

descriptively. To determine intra-rater reliability for cleft width measurement in Study I, III, and IV and inter-reliability between speech ratings from the medical records and the external listener in Study I, the single measures intra-class correlation coefficient with a 2-way mixed-effects model (ICC) was used. Statistical analyses were performed in IBM SPSS Statistics, version 23 or 29, or in R. The level of significance was set at $\alpha < 0.05$ (2-tailed). The tests used for statistical analyses are presented in Table 4.

Table 4. Test used for statistical analyses in the studies.

Test	Study I	Study II	Study III	Study IV
Logistic regression	X		X	X
Linear regression				X
Ordinal regression			X	X
ICC	X		X	X
Chi2 test	X	X		X
Kruskal-Wallis test	X		X	
Fishers exact test		X		
Mann-Whitney U test		X		
Spearman's rank order correlation test			X	

ICC = Intra-class correlation coefficient

Reliability

Study I

Intra-rater reliability was calculated using the single measures intra-class correlation coefficient with a 2-way mixed-effects model (ICC). The levels of observed agreement were interpreted according to Cicchetti (89) as follows: $<.40$ is poor, $.40-.59$ is fair, $.60-.74$ is good, and $.75-1.00$ is excellent. Intra-rater reliability was excellent for hypernasality, audible nasal emission (0.96, 95% CI 0.90-0.99 and 0.97, 95% CI 0.90-0.99), and measurement of cleft width (0.96, 95% CI 0.92-0.98). For glottal articulation, the agreement was 100%. The reliability between the external SLP and medical records was fair for hypernasality and audible nasal emission (0.51, 95% CI 0.31-0.67 and 0.51, 95% CI 0.30-0.67 respectively) and 100% for glottal articulation.

Study II

Inter- and intra-rater agreement was calculated using point-by-point comparisons for correctly articulated target sounds, VPI symptoms, and non-oral articulation. The frequency of 1) agreement between all three raters, 2) agreement between two of the three raters, and 3) no agreement was used to calculate inter-rater agreement for hypernasality and VPC-R. The intra-rater agreement for both hypernasality and VPC-R was calculated using point-by-point

agreement and reported in percent. Table 5 and 6 provide information on inter- and intra-rater agreement on speech outcomes.

Study III and IV

The single measures intra-class correlation coefficient with a 2-way mixed-effects model (ICC) showed that intra-rater reliability for cleft width measurement in Study III and IV was good (.686, 95% CI .315-.874 and .718, 95% CI .475-.860 respectively) when interpreted according to Cicchetti (89). Inter- and intra-rater agreement for correctly articulated target sounds, manner, place, VPI symptoms, and non-oral articulation were calculated using point-by-point comparisons. The frequency of 1) agreement between all three raters, 2) agreement between two of the three raters, and 3) no agreement was used to calculate interrater reliability for hypernasality and VPC-R. Intra-rater agreement for both hypernasality and VPC-R was calculated using point-by-point agreement and reported in percent. Table 7 and 8 provide information on inter- and intra-rater agreement on speech outcomes.

Table 5. Inter- and intra-transcriber agreement for the transcriptions in Study II at word and sentence levels at five and ten years of age. Median percentages (min-max).

	Inter-transcriber agreement		Intra-transcriber agreement	
	Word level	Sentence level	Word level	Sentence level
Five years				
Correct/incorrect articulation	88% (69-98)	89% (71-100)	95% (84-100)	91% (83-100)
Symptoms of VPI	85% (49-97)	77% (47-100)	95% (81-100)	87% (74-100)
Presence of non-oral articulation	96% (76-100)	95% (74-100)	98% (82-100)	99% (83-100)
Ten years				
Correct/incorrect articulation	97% (73-100)	97% (74-100)	98% (85-100)	100% (89-100)
Symptoms of VPI	95% (56-100)	97% (54-100)	92% (74-100)	94% (74-100)
Presence of non-oral articulation	100% (90-100)	100% (100)	100% (95-100)	100% (97-100)

VPI = velopharyngeal insufficiency

Table 6. Inter- and intra-rater agreement in Study II for hypernasality and VPC-R at five and ten years of age.

	Hypernasality		VPC-R	
	Five years	Ten years	Five years	Ten years
Inter-rater				
All three raters agree (%)	38%	48%	65%	77%
At least two out of three raters agree (%)	97%	98%	100%	100%
No agreement (%)	3%	2%	0%	0%
Intra-rater				
Rater 1 (%)	50%	93%	93%	93%
Rater 2 (%)	79%	64%	93%	93%
Rater 3 (%)	71%	93%	93%	86%

VPC = velopharyngeal competence

Table 7. Inter- and intra-transcriber agreements for transcriptions in Study III and IV. Median percentages (min-max).

Agreement on	Inter-transcriber agreement		
	Rater 2	Rater 3	Rater 3
Correct/incorrect articulation	91% (65-100)	95% (85-100)	97% (84-100)
Manner of articulation	98% (76-100)	98% (88-100)	100% (96-100)
Place of articulation	90% (74-100)	95% (77-100)	96% (83-100)
Symptoms of VPI	93% (70-100)	97% (87-100)	97% (86-100)
Presence of non-oral articulation	100% (82-100)	100% (87-100)	100% (89-100)
Correct/incorrect articulation	91% (58-100)	96% (81-100)	97% (84-100)
Manner of articulation	98% (76-100)	98% (88-100)	100% (96-100)
Place of articulation	89% (65-100)	95% (77-100)	97% (83-100)
Symptoms of VPI	93% (70-100)	97% (79-100)	96% (83-100)
Presence of non-oral articulation	100% (70-100)	100% (85-100)	100% (89-100)

VPI = velopharyngeal insufficiency.

Table 8. Inter- and intra-agreement for ratings of hypernasality and VPC-R in Study III and IV.

Inter-rater agreement	Study III		Study IV	
	Hypernasality	VPC-R	Hypernasality	Hypernasality
All three raters agree (%)	48%	58%	45%	45%
At least two out of three raters agree (%)	92%	98%	92%	92%
No agreement (%)	8%	1%	8%	8%
Rater 1	80%	74%	77%	77%
Rater 2	85%	95%	85%	85%
Rater 3	70%	100%	69%	69%

VPC = velopharyngeal competence.

Results

Study I

Of the 213 participants, 23 individuals (11%) required secondary palatal surgery due to dehiscence. Among these 23 patients, nine had a pharyngeal flap at the time of the salvage palatoplasty, while another nine required speech-improving surgery later on. A further 41 patients required speech-improving surgery, making a total of 50 patients (23%) in the whole cohort that required speech-improving surgery. In addition, 28 patients (13%) underwent fistula repair.

Cleft width was significantly associated with the rate of secondary surgery due to dehiscence, but cleft type was not. After excluding those who underwent secondary surgery due to dehiscence, the logistic regression analysis included 190 individuals, revealing a significant association between cleft width but not cleft type and the rate of speech-improving surgery; see Table 9.

Table 9. Logistic regression analysis showing the association between cleft width, cleft type, and the rate of secondary palatal surgery.

Variable	Secondary palatal surgery due to dehiscence of soft palate			Speech-improving surgery		
	OR	95% CI	<i>p</i>	OR	95% CI	<i>p</i>
UCLP	Ref		.152	Ref		.920
BCLP	.373	.088-1.585	.182	.854	.320-2.78	.752
SHCP	1.733	.625-4.800	.290	1.074	.466-2.479	.867
Cleft width	1.102	1.041-1.167	<.001	1.058	1.010-1.107	.017

UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, OR = odds ratio, CI = confidence interval.

One hundred and forty individuals had at least three out of four possible speech assessments and were included in the speech outcome analysis (61 with UCLP, 20 with BCLP, 59 with SHCP). Seventy-three had fewer than three assessments due to difficulty participating, technical failure during recording, or because the assessment was not conducted on time (3 years, +/-3 months; 5 years, +/- 6 months; 10 +/- 1 year; and 16 years, +/- 1 year). Five of those included in the speech analyses were diagnosed with a syndrome (three with UCLP and two with SHCP with syndromic Robin sequence), and nine had non-syndromic Robin sequence. Individuals who had secondary surgery due to fistulas or dehiscence were included in the speech analyses. In contrast,

individuals who had undergone speech-improving surgery at the time of the routine control at any age were excluded from the analyses of speech variables from that age and onward.

No significant differences between cleft types were found for hypernasality, audible nasal emission or glottal articulation at any age (hypernasality; 3 years: $\chi^2(6) = 3.16, p = .788$; 5 years: $\chi^2(4) = .42, p = .981$; 10 years: $\chi^2(2) = .26, p = .876$; 16 years: $\chi^2(4) = 6.02, p = .197$, audible nasal emission; 3 years: $\chi^2(4) = .31, p = .989$; 5 years: $\chi^2(4) = 7.75, p = .101$; 10 years: $\chi^2(4) = 2.54, p = .637$; 16 years: $\chi^2(6) = 7.18, p = .305$, glottal articulation; 3 years: $\chi^2(2) = 3.11, p = .212$; 5 years: $\chi^2(2) = 2.93, p = .231$; 10 years: $\chi^2(2) = 2.51, p = .286$; 16 years: no glottal articulation was noted).

The distribution of cleft width between the scale steps differed significantly for both hypernasality (Figure 6) and audible nasal emission (Figure 7) at age five but not at other ages.

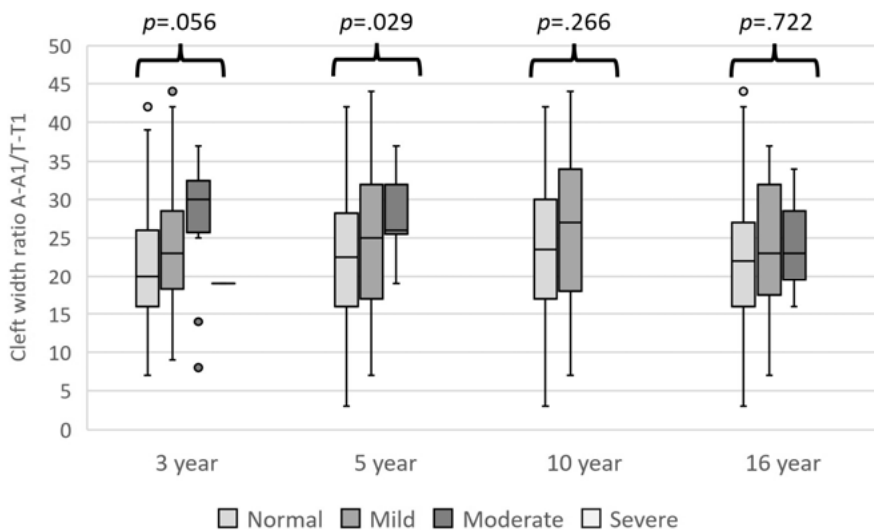


Figure 6. Median cleft width distributed on the scale steps for hypernasality at the respective follow-up time points.

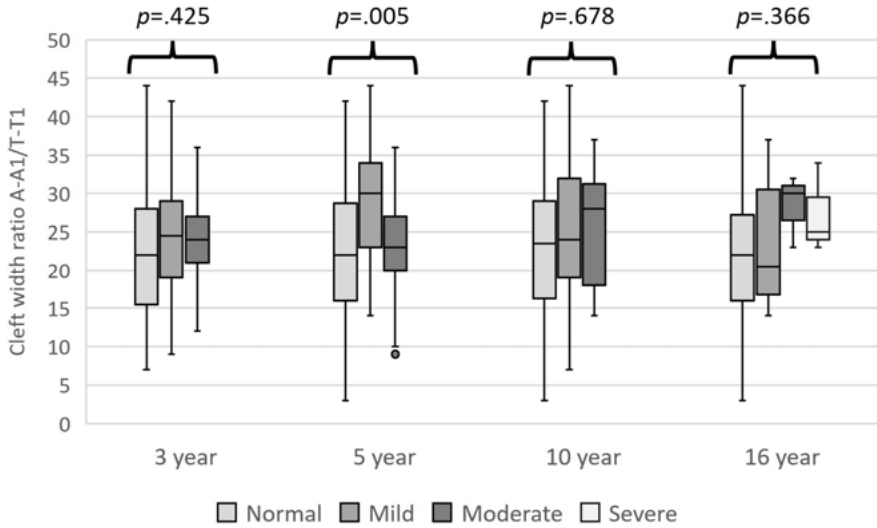


Figure 7. Median cleft width distributed on the scale steps for audible nasal emission at the respective follow-up time points.

At ages three and five, cleft width differed significantly between those with presence and absence of glottal articulation. It did not differ at age ten, and at age 16, no one had glottal articulation; see Figure 8.

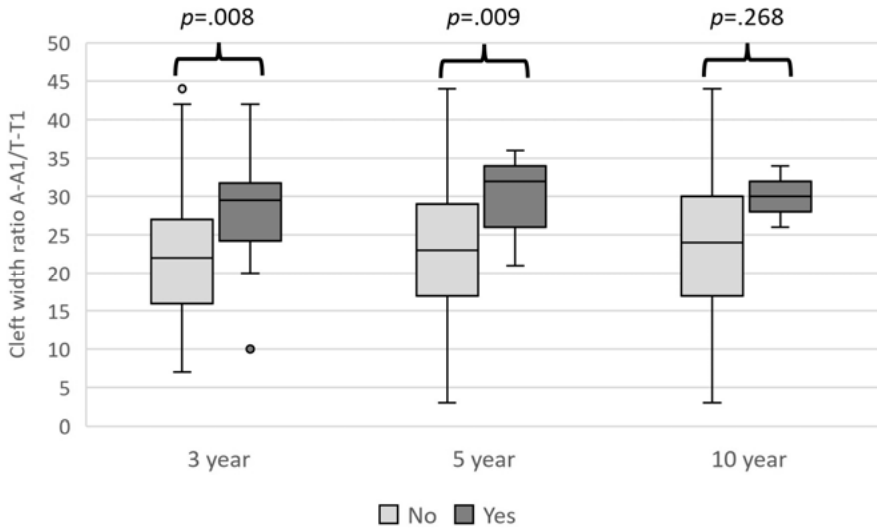


Figure 8. Median cleft width distributed on the scale steps for the presence and absence of glottal articulation at the respective follow-up time points. No one had glottal articulation at age 16.

Study II

When comparing internationally adopted (IA) children to non-adopted (NA) children, a significant difference was observed at the age of five using VPC-Sum ($\chi^2(2) p = .031$), but not using VPC-R ($\chi^2(2) p = .492$). Similarly, at the age of ten, a significant difference was observed using VPC-Sum ($\chi^2(2) p = .003$) but not using VPC-R ($\chi^2(2) p = .264$). At both ages, fewer children in the IA group had competent VPF than did in the NA group, regardless of measurement. The number of children with incompetent VPF differed somewhat between the composite variable VPC-Sum (IA $n = 6$, NA $n = 7$) and the rating VPC-R (IA $n = 7$, NA $n = 5$).

Median PCC at the word level did not differ significantly between IA children (68%) and NA children (83%) at age five ($U = 180.00$, $z = -1.86$, $p = .063$), but it did at age ten. Median PCC was 90% in the IA group compared to 98% in the NA group ($U = 164.50$, $z = -2.27$, $p = .023$). At the sentence level, the situation was reversed with a significant difference ($U = 156.50$, $z = -2.38$, $p = .017$) at age five (IA 60%, NA 89%) but a non-significant difference at age ten ($U = 212.50$, $z = -1.25$, $p = .212$) (IA 97%, NA 100%); see Figure 9.

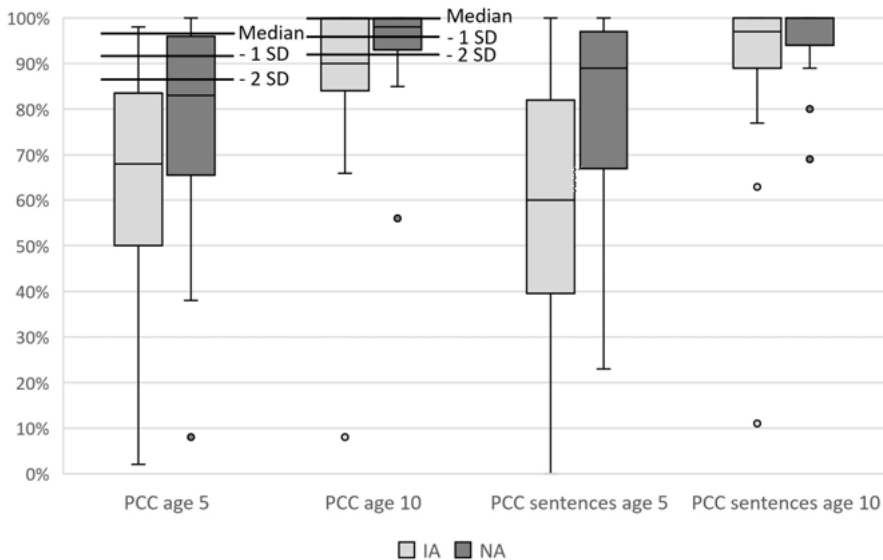


Figure 9. Median percent consonants correct (PCC) at word level and sentence level at ages five and ten years for internationally adopted (IA) and non-adopted (NA) children. The norm PCC values at word level for 5- and 10-year-old Swedish speakers without cleft palate at the median, - 1 SD, and - 2 SD are indicated by horizontal lines. SD = standard deviation.

Both CSCs, DSCs, and s-errors were more frequent in IA children (78%, 39%, and 39%, respectively) than in NA children (48%, 22%, and 26%,

respectively) at age five, but not to a significant degree. An in-depth analysis of subtypes of articulation error subsumed under CSCs and DSCs showed that all errors except active nasal fricative were more frequent in the IA than in the NA group. The difference was significant for glottal reinforcement or substitution ($p = .029$) but not for any other articulation error. At ten years of age, 48% of the IA children and 13% of the NA children still presented with CSCs ($p = .023$). Oral retracted articulation remained in 26% of the IA and 9% of the NA children. Non-oral articulation was present in 22% of the IA children compared to 4% of the NA children. S-errors were found in 22% of the IA children compared to 9% of the NA children. At ten years of age, DSCs had ceased in all but two IA children who still had minor difficulties, one with voicing errors and one with stopping.

Thirty-nine percent of the IA children had, by the age of ten, received speech-improving surgery compared to 26% of the NA children. The difference was not statistically significant ($p = .530$).

At five years of age, the IA children had an average of 19 visits to a speech-language pathologist (SLP), while NA children had 14 visits on average. IA children had an average of 12.5 visits between the ages of five and ten, while NA children had eight. The differences between the groups were not statistically significant at any age ($p = .450$ and $.335$ respectively).

Study III

As shown in Table 10, the rate of secondary palatal surgery due to dehiscence, fistula, or VPI was significantly associated with cleft width but not with the surgical technique.

Table 10. Logistic regression analysis on how surgical technique and cleft width were associated with the rate of secondary palatal surgery due to dehiscence, fistula, or velopharyngeal insufficiency (VPI).

Variable	OR	95% CI	<i>p</i>
Non-dissection (n=16)	REF		.361
Transition (n=14)	.659	.106-4.093	.655
Sommerlad (n=32)	.309	.059-1.621	.165
Cleft width	1.141	1.021-1.275	.020

OR = odds ratio, CI = confidence interval.

Primary VPI can be used to represent the results of the surgical techniques used accurately (33, 90). Primary VPI combines individuals with a VPC-R score = 2 or a VPC-Sum score ≥ 4 based on the SLPs' assessment with those who underwent speech-improving surgery before age five, thus representing the results of the primary palatoplasty. In the ordinal regression analyses, primary VPI was used, i.e., those who had speech-improving surgery were

counted as having VPI regardless of the assessments and were consequently put into the group with a VPC-Sum score ≥ 4 (transferred into VPC-Sum = 2 in the analyses) and VPC-R = 2. Surgical technique was not significantly associated with VPC-R, but cleft width was. Neither surgical technique nor cleft width was associated with VPC-Sum; see Table 11.

Table 11. Ordinal regression analyses on how surgical technique and cleft width were associated with VPC-R and VPC-Sum.

Variable	VPC-R			VPC-Sum		
	OR	95% CI	<i>p</i>	OR	95% CI	<i>p</i>
Non-dissection (n=16)	REF			REF		
Transition (n=14)	3.448	.663-17.932	.141	3.784	.731-19.585	.113
Sommerlad (n=32)	.814	.164-4.039	.801	1.227	.270-5.566	.791
Cleft width	2.700	1.053-6.919	.039	1.985	.845-4.662	.116

OR = odds ratio, CI = confidence interval.

There was no significant correlation between cleft width and PCC ($\rho = -.128, p = .329$), and there were no differences in PCC between surgical techniques ($H(2) = .070, p = .966$). Results divided by technique for the surgeon active throughout the study period are displayed in Figure 10 a-c.

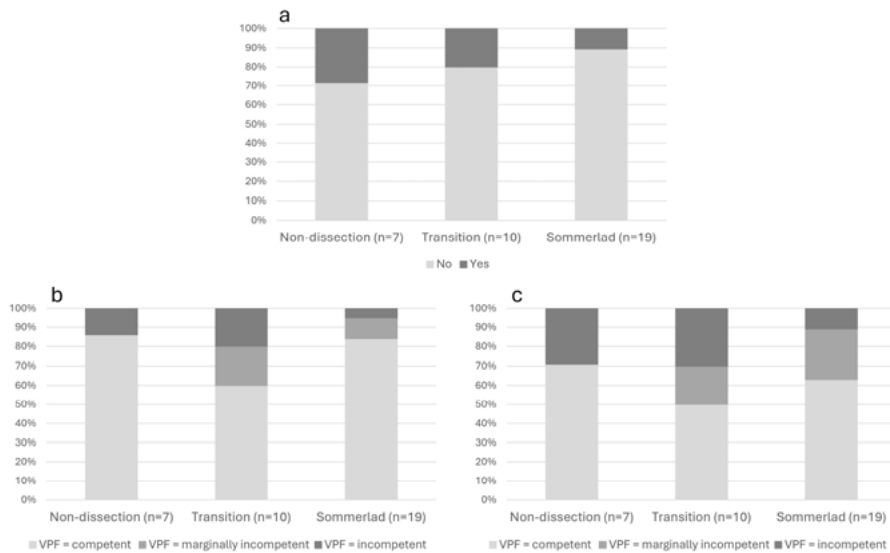


Figure 10. a) The rate of secondary palatal surgery, b) Velopharyngeal function (VPF) assessed with VPC-R, and c) VPF assessed with VPC-Sum divided into surgical techniques for the surgeon active throughout the study period. Those who had undergone speech-improving surgery before the assessment are counted as having velopharyngeal insufficiency.

Study IV

Cleft type was not associated with the rate of secondary surgery due to dehiscence or fistula (UCLP versus BCLP, OR 1.507, 95% CI: .336-6.760, $p = .593$, UCLP versus SHCP, OR 2.435, 95% CI: .535-11.079, $p = .250$) or with the rate of speech-improving surgery (UCLP versus BCLP, OR 1.515, 95% CI: .322-7.131, $p = .599$, UCLP versus SHCP, OR 4.858, 95% CI: .950-24.850, $p = .058$). Cleft width was also not associated with these rates (OR 1.033, 95% CI: .952-1.122, $p = .437$ and OR 1.099, 95% CI: .998-1.209, $p = .054$ respectively). Table 12 shows that cleft width but not cleft type was significantly associated with PCC and PCC-A. Median PCC and PCC-A divided according to cleft type are presented in Figure 11.

Table 12. Linear regression analysis on how cleft type and cleft width were associated with the percent of consonants correct (PCC) and PCC adjusted for age (PCC-A).

Variable	PCC			PCC-A		
	OR	95% CI	p	OR	95% CI	p
UCLP (n=62)	REF			REF		
BCLP (n=17)	-3.488	-33.340-6.363	.483	-3.031	-12.945-6.882	.545
SHCP (n=21)	-3.389	-13.769-5.993	.436	-4.568	-14.511-5.374	.364
Cleft width	-0.676	-1.172- -0.179	<.01	-0.604	-1.104- -0.105	<.05

UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, OR = odds ratio, CI = confidence interval.

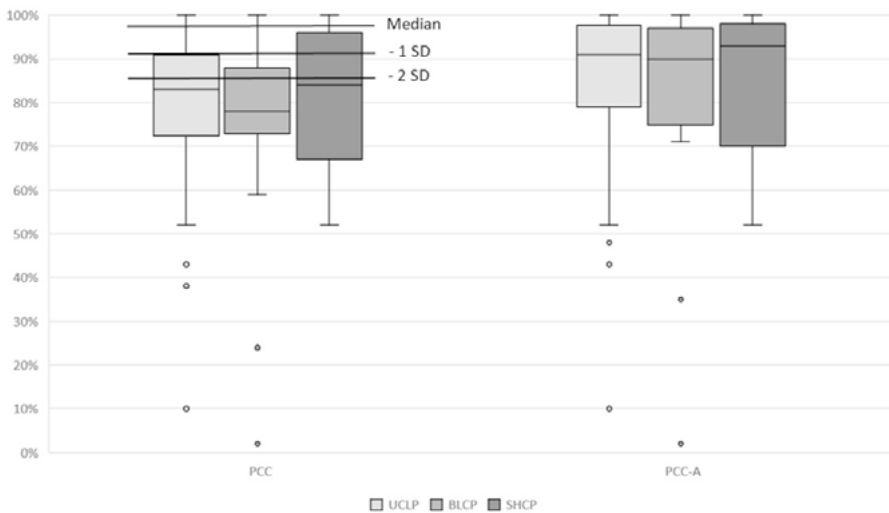


Figure 11. Comparison of median percent of correct consonant (PCC) and PCC adjusted for age (PCC-A) between cleft types at five years of age. The norm PCC values for 5-year-old Swedish speakers without cleft palate at the median, - 1 SD, and - 2 SD are indicated by horizontal lines. SD = standard deviation, UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = cleft in the soft and hard palate.

The BCLP group had the lowest PCC and PCC-A at 78% and 90%, followed by 83% and 91% in the SHCP group and 84% and 93% in the UCLP group.

The occurrence of CSCs did not differ significantly between cleft types ($\chi^2(2) p = .869$) but was most frequent in the BCLP group (65%), followed by the UCLP group (58%) and the SHCP group (57%). Individuals with BCLP more commonly had oral and non-oral articulation errors (59% and 24%, respectively) compared to those with UCLP (52% and 15%, respectively) and SHCP (48% and 14%, respectively).

The incidence of DSCs did not differ significantly between cleft types ($\chi^2(2) p = .531$) but was most frequent in the BCLP group (65%), followed by 57% in the SHCP group and 50% in the UCLP group. When examining different types of DSCs, it was found that velar fronting was a rare occurrence. However, it was more common in individuals with SHCP (10%) than those with BCLP and UCLP (0% and 3%, respectively). Both stopping and voicing errors were about equally frequent in all groups (UCLP 11% and 10% respectively, BCLP 12% and 12% respectively, SHCP 14% and 14% respectively).

Having both CSCs and DSCs was most common in the SHCP and BCLP groups (48% and 47%, respectively) compared to the UCLP group (27%). S-errors were present in 68% of the UCLP group, 65% of the BCLP group, and 57% of the SHCP group, and /s/ was the most affected target sound in all groups.

Neither cleft type (UCLP versus BCLP, OR 2.651, 95% CI: .941-7.470, $p = .065$, UCLP versus SHCP, OR 2.402, 95% CI: 0.790-7.301, $p = .122$) nor cleft width (OR 1.332, 95% CI: .755-2.351, $p = 1.332$) was associated with VPC-Sum.

Discussion

In the present doctoral project on cleft lip and palate, the influence of cleft width, cleft type, surgical technique, and surgeon experience, believed to impact surgical and speech outcomes, were explored. Cleft width turned out to be the only independent predictor of the outcomes included in the present thesis. A comparison between internationally adopted and non-adopted children at five and ten years of age was also conducted, showing that internationally adopted children are at higher risk of persistent articulation difficulties.

The results from Study I, III, and IV combined indicate a strong association between cleft width and the rate of speech-improving surgery. Additionally, there was a significant association between cleft width and the rate of secondary palatal surgery due to fistula or dehiscence in Study I. In Study II, cleft width could not be taken into consideration since the adopted children had either already undergone surgery prior to being adopted or had their first palatal surgery on average at two years of age, which meant they were not comparable to the non-adopted children.

Although some differences were observed, neither cleft type nor the surgical technique used proved to be indicative of the rate of secondary palatal surgery. Study III showed that the rate of primary VPI increased during the transition period when a more extensive dissection of the levator veli palatini muscle was introduced. However, the rate of speech-improving surgery was similar between the techniques. The total rate of secondary palatal surgery decreased from 25% in the non-dissection group to 16% in the Sommerlad group, indicating better outcomes at five years of age in the latter group. However, we know from Study I and II that speech-improving surgery may be performed after age five. Indeed, 34% of the individuals requiring speech-improving surgery in Study I underwent surgery after seven years, and 53% of the individuals in Study II who underwent surgery did so between five and ten years of age. Additionally, between routine controls at ages ten and 16, some individuals in Study I developed VPI, underlining the importance of routine controls and longitudinal studies covering development until adulthood. In Study II, no statistically significant difference was found between adopted and non-adopted children regarding speech-improving surgery. However, 39% of the internationally adopted children, compared to 26% of the non-adopted children, had undergone speech-improving surgery by the age of

ten. This suggests that internationally adopted children are at higher risk of VPI, which is probably partially due to their older age at primary surgery.

Signs of VPI include hypernasality, audible nasal emission, weak pressure articulation, and non-oral articulation. Considered separately, the individual variables do not provide an overall picture of VPF, and in recent studies, an overall assessment of VPF, such as VPC-R, or a calculation of the sum of ratings, such as in VPC-Sum has been used (20, 24, 33). Throughout the 29-year duration of assessments, from which the data in Study I was collected, the speech assessment routine and materials utilized were changed. During the earlier years included in the study, weak pressure consonants were not rated or described, and only glottal articulation was rated, while other non-oral articulation errors were inconsistently described and never rated. This made it impossible to convert the ratings into a composite score representing the overall rating of VPF. As a result, conducting a regression analysis to investigate the association with VPF, which was done in Study III and IV, was impossible. Nevertheless, it was evident that wider clefts had more indications of VPI, particularly during the preschool years. In Study III, the results partially supported the findings of Study I. Specifically, cleft width was significantly associated with VPF when using VPC-R. It is worth noting, however, that cleft width did not show any significant association with VPF when using VPC-Sum in either Study III or IV. However, individuals with VPI had a wider median cleft width than those with competent or marginally incompetent VPF.

In Study I and IV, no association was found between cleft type and variables associated with VPF or VPF assessed with VPC-Sum. There was also no association between surgical technique and VPF in Study III. This may be due to the small sample sizes, especially in Study III. Further division of the already small sample sizes into different groups may result in the loss of possible associations.

As a group, most children in Study II-IV had lower PCC than the norms for typically developed children without CLP (4), regardless of cleft type or whether they were adopted. These results confirm findings from previous similar studies. (62, 67, 69, 91). In fact, the medians for the respective cleft types and both adopted and non-adopted children were below -2 SD, which indicates severe difficulties with articulation proficiency. It also became apparent that children with CLP not only exhibit CSCs but also frequently exhibit DSCs, at least at five years of age. DSCs, also known as phonological errors, are more prevalent in children with cleft palate than those without (63, 92, 93). According to Lien et al. (94), phonological errors were found to be the most common speech errors in a group of children with cleft palate aged four to seven years. In Study II and IV, DSCs were common, but the most prevalent articulation error in both studies was oral CSCs. Lien et al. (94) also found that phonological errors decreased significantly with age, which was not observed for CSCs. Their findings are consistent with those of Study II, which showed that DSCs were frequent when the children were five years old but had resolved in all

but two adoptees by age ten. At age ten, both groups showed persistent CSCs, with 48% of adoptees and 13% of non-adoptees affected.

In Study II, at age five, 52% of non-adopted and 30% of adopted children had non-oral articulation errors, particularly glottal articulation. Even at the age of ten, 22% of adopted children and 4% of non-adopted children continued to have non-oral articulation errors, primarily glottal articulation. This indicates that non-oral articulation errors are challenging to correct even after restoring velopharyngeal function and/or speech intervention. As glottal articulation severely impairs intelligibility, this must inevitably affect the individual's ability to communicate.

It is essential to consider that several factors distinguish internationally adopted children from non-adopted children, including their circumstances during the first years of life. For instance, the majority of adopted children spend their early years in orphanages, where the quality of care and stimulation may differ significantly from that provided within a family. Children at institutions are at risk for different health and developmental issues (95) and have a higher prevalence of speech and/or language impairment (96). Studies have shown that internationally adopted children without CLP reach an age-appropriate language and articulation proficiency level within two or three years after adoption (97-99). In contrast, studies investigating language ability in school-aged children without CLP show a somewhat different picture, with internationally adopted children without cleft palate scoring below the norms (100-102). The results of the above studies (100-102) were supported by a Swedish study in which internationally adopted children with and without cleft palate were compared at seven to eight years of age. Both groups scored lower than normative and reference values on language measures. However, only the cleft palate group scored lower on articulation proficiency (103). Findings showing that the articulation ability of adopted children without CLP is age-appropriate (99, 103) may lead to the conclusion that the language switch does not affect articulation ability. However, children with cleft lip and palate are at risk of poor articulation development, and internationally adopted children are at an even greater risk. As a result, the language switch may further add to their vulnerability to articulation difficulties. Additionally, none of the adopted children in our study were fitted with tympanostomy tubes to aid hearing prior to adoption, which could have potentially affected their early speech and language development. Moreover, children who are adopted usually have surgery later than non-adopted children. This delayed surgery may be a contributing factor in explaining why a larger number of adopted children continue to have articulation difficulties well into their school years. By the time surgery is performed, the manner in which the child produces speech sounds may already be established, making pronunciation more challenging to correct.

In Study IV, cleft width but not cleft type was associated with PCC and PCC-A. The significant differences in articulation proficiency between cleft

types demonstrated in previous studies (33, 64, 67-70, 91) were not confirmed in our research. However, we did find that children with BCLP had poorer articulation, consistent with previous research.

Taken together, /s/ was the most common target sound affected by deviation. S-errors at age five are considered age-appropriate exhibited in ~30% of Swedish-speaking children without cleft palate (4). In Study II, 26% of the non-adopted children had s-errors at five years of age and were, in that respect, comparable to children without cleft palate. However, in Study IV, s-errors were present in 52-65% of the children. It is possible that the difference observed between the outcomes of the two studies can be explained by the fact that more children in Study II had non-oral errors and that /s/ produced non-orally was not categorized as an s-error but as a CSC. However, the difference could also be attributed to discrepancies in the transcriptions, as different transcribers were used in the two studies. S-errors might be considered less severe than, for example, stopping or velarization, but they can still affect how others perceive a person's speech. Even minor articulation deviations get noticed and commented on by peers, while minor signs of VPI are disregarded (104, 105). This highlights the importance of also considering minor speech deviations when planning speech intervention.

Methodological considerations

The sample sizes were relatively small, especially in Study II and III. Dividing an already small group into smaller groups may lead to type I or type II errors, which must be kept in mind when interpreting the results.

The decision to include children with syndromes and/or Robin sequence (RS) in Study I was based on the fact that with only a very small proportion of diagnosed children, we suspected that genetic syndromes were underdiagnosed in this historical cohort. The reported incidence of syndromes and/or additional malformations in European children with cleft lip with or without cleft palate is about 30%, and the corresponding figure is approximately 50% in children with cleft palate without cleft lip (106, 107). In the total cohort of 313 individuals, 11.5% were diagnosed with a syndrome or additional malformation, and the same proportion were included in the study: 3% in the UCLP group, 11% in the BCLP group, and 17% in the SHCP group had a diagnosis of syndrome and /or RS. This suggests underdiagnosis. In research, grouping children with syndromes or additional malformations into separate categories and conducting sub-analyses is common. However, we decided not to exclude them or to perform a sub-analysis, as this approach would still likely include many children with syndromes or malformations in the "non-syndromic" group, potentially leading to inaccurate results.

To be able to compare surgical techniques, rather large groups are needed to detect possible differences. Although we have the second largest unit in

Sweden, the population in our region is still small, and we need to include participants over many years to gather sufficient cohorts. As the study period is long, it should be noted that multiple surgeons were involved in Study III, which may be a confounding factor. One of the surgeons was consistently active throughout the entire study period, making it possible to compare techniques with the surgeon being constant. However, this analysis may have been affected by the increasing experience of that particular surgeon and, thus, perhaps not reflecting differences between surgical techniques.

Standardized and recorded speech assessments using standardized speech material at routine controls enable us to conduct studies based on the recordings made at these routine controls. Admittedly, the content and sometimes the quality of the collected material is beyond our control, but we can use material collected during long periods of time and perhaps increase the included cohorts. The material also enables cross-sectional analyses like those carried out in Study II-IV. Re-assessments based on recordings ensure that assessments are made within a short time frame with consistent definitions and rules, consequently reducing the risk of a changed understanding of the definitions and rules. However, the drawback of using data not initially intended for research purposes is that we need to adapt our research inquiries to the information gathered. One advantage of this method is that it does not require additional resources during data collection. The data are collected based on clinical necessity, which means no extra burden is imposed on the study subjects, as might be the case in a prospective study. Nevertheless, a prospective study has the benefit of enabling us to tailor the data we collect to the specific research questions.

As we did not have a prospective approach in the present thesis, we could not investigate other aspects of speech development that might have an influence on articulation ability. Meta-phonological ability is such an aspect as phonological difficulties are common in children with CLP (63, 92-94), and further research on speech input processing is required (108). Another area of interest is language ability, as children with cleft palate have been shown to have poorer language ability (109) and a higher risk of language impairment (110).

Regardless of study design, inter- and intra-rater reliability is crucial when interpreting results from speech assessments. Several factors may influence reliability, and it is well-known that high inter-rater reliability for different variables, not least hypernasality, is hard to achieve (111).

The listener's experience is crucial as more experience increases reliability (112). All raters who participated in the present project had adequate experience of cleft palate speech. However, Kreiman et al. (113) proposed that listeners develop an individualized internal standard, which can vary among listeners and may be inconsistent over time, regardless of the listener's experience. Variable definitions, attention lapses, and poor audio quality may also influence assessments.

One way to enhance inter- and intra-rater reliability is to perform training/calibration sessions, which has proven to increase agreement between raters (51, 55, 57, 58, 114). Calibration sessions were conducted before carrying out assessments in Study II-IV, and the definitions of the included variables and scale steps were thoroughly discussed. When the transcriptions for Study II were finalized, it was discovered that the main transcriber had only used the sign for alveolo-palatal fricative (ɛ) and never the sign for palatal fricative (ç) while transcribing /s/, and thus, these transcriptions could not be divided into CSCs and DSCs. As a result, there was a comprehensive discussion on transcribing different realizations of /s/ in the calibration session for Study III and IV. This allowed us to divide s-errors into CSCs and DSCs in Study IV, which enhanced the level of detail compared to Study II. Still, the reliability was not flawless and could perhaps have been improved through additional calibration.

For Study I, we could not re-assess all the material and had to rely on the assessments from the original evaluation retrieved from the medical records. Thus, the retrospective nature of Study I compromises the reliability of the results. As the assessments are made over a long time period, definitions of how to assess different speech variables may have changed. Indeed, the ordinal scale changed from a 5-point to a 4-point one, and glottal articulation went from being rated on an ordinal scale to being based on transcriptions and reported in number of occurrences. In total, five SLPs were involved during the study period. When clinicians work together, they tend to develop a consistent internal standard by listening to the same material and discussing individuals, which helps them reach consensus on specific aspects of their work, such as hypernasality (114). However, given that the assessment period covers almost three decades, the internal standard may have changed over time. In an effort to increase reliability, an external SLP did re-assessments from audio recordings for ten percent of the whole material. The reliability between the medical records and the SLP assessments was only fair for both hypernasality and nasal emission. As for hypernasality, the level of reliability was comparable to several other studies (33, 67, 115), but for audible nasal emission, it was somewhat lower than expected (116, 117). Comparisons between ratings of hypernasality based on audio recordings and the original clinical evaluation have shown good intra-rater reliability, comparable to intra-rater reliability based on assessments and re-assessments from audio recordings. However, when comparing the reliability between assessments of audible nasal emission based on audio recordings and the original clinical evaluation, intra-rater reliability was poor (118). The authors concluded that ratings of audible nasal emission from audio recordings may not be a valid measure of VPI. The results of Williams et al. (118) may help partly explain our low reliability between the re-assessment from audio recordings and the original assessments for audible nasal emission. Poor sound quality may also account for differences between ratings from the clinical assessment and that based on audio recordings, and it

may be that audible nasal emission is more vulnerable to poorer sound quality than hypernasality. In addition, different SLPs from different CLP teams, probably with different internal standards and perhaps with varying definitions of scale steps, may have contributed to the assessments' variability.

In the present project, we set the criterion for the presence of an articulation error as three or more occurrences. This cutoff has been used in the Scandleft studies (16, 17, 51, 119), from which the present project's definitions are taken. However, other cutoffs have been used; for instance, a cutoff of one has been used in studies originating from the United Kingdom (33, 64). The cutoff of one error may seem too harsh, but in the studies mentioned above (33, 64), the speech was evaluated by at least two SLPs, and consensus was reached, reducing the risk of mishearing. Still, it is important to note that a single instance of mispronunciation may not necessarily indicate a child's difficulty with articulation but can simply be a slip of the tongue. It has been recommended that more than one occurrence of an error should be observed to conclude its presence (120). In cases where consensus listening is not employed, such as in the present thesis, there is a higher risk of mishearing, and a single occurrence of an error may not be enough to determine whether a type of error actually exists. Conversely, allowing for three errors may be too generous, and it may be more appropriate to allow for two errors – striking a balance between reducing the risk of misinterpretation and maintaining reliability.

At the beginning of the present project, the ambition was to include continuous speech, preferably conversational speech, to provide further insight into the children's VPF, articulation ability, and intelligibility. It became apparent during the process of editing audio recordings that continuous speech was missing in many cases at both five and ten years of age. In some cases, the SLP attempted to elicit continuous speech through a brief conversation with the child or by asking the child to retell the Bus Story (121) or to describe a picture. However, it was quite common for the segments of continuous speech to be very short; sometimes, it was obvious that the children did not want to or could not participate in more conversation/storytelling, but occasionally, the recording was interrupted by the SLP, perhaps due to time limitations. Reluctance or inability to participate in longer sections of continuous speech may stem from, for example, shyness, awareness of having an unintelligible speech, language difficulties, or tiredness. Nevertheless, we should strive to elicit and record longer sections of continuous speech and, preferably, conversational speech, as this enables us to assess intelligibility, further analyze articulation ability and VPF, and possibly determine the level of speech functionality.

When conducting studies on speech outcomes in children with cleft palate, it is desirable to report information on hearing. Otitis media with effusion associated with conductive hearing impairment is common in this group (122) and could be a confounding variable. Our clinic has made efforts to implement routine hearing tests as part of the routine visit, but they have not yet been

conducted due to limited resources. This does not mean that the children's hearing is not being controlled, but as we care for children from many different regions, we do not have immediate access to the medical data. The relationship between hearing impairment and consonant proficiency is currently not well understood, with some studies reporting a significant association (33, 123, 124) while others contradict this finding (63, 68). Nevertheless, the lack of information on hearing acuity is a weakness throughout the present thesis, and such information would have been valuable information in determining possible predictors of speech outcomes.

Clinical implications

It is clear that children with CLP face articulation difficulties and require speech therapy. Study II and IV show that treatment should address both errors related to the cleft and those related to development. Moreover, Study II indicates that the amount of intervention varies across children and that earlier and increased intervention is probably necessary to address articulation difficulties.

Several factors influence speech development, articulation proficiency, and velopharyngeal function. In our research, we found that cleft width is a robust factor that can predict outcomes. This finding may serve as a useful prognostic tool in clinical practice.

During the transition period in Study III, it became evident that the primary VPI increased after the introduction of radical muscle dissection. However, after the proper technique, as described by Sommerlad, was introduced, there was an instant improvement. Although the surgeon's enhanced experience may have contributed to the improvement, it is also possibly due to the systematic implementation following calibration with Brian Sommerlad. This highlights the significance of introducing new surgical techniques in an organized and systematic manner.

Two areas have been identified as needing improvement based on the experiences gained in the present studies. First, the lack of information on the hearing status of a child not only poses a problem during studies, but it is also crucial in interpreting clinical evaluations and planning interventions. The second area for improvement is the recording of continuous speech. Although clinicians hear more speech during a visit and can make a valid assessment of intelligibility, for future studies, it would be desirable to record continuous speech. Such recordings reveal other aspects of a child's articulation proficiency and can serve as an additional sample when evaluating intervention.

Future perspectives

It is essential to consider long-term outcomes before making any definitive conclusions about factors that may impact different outcomes, not least surgical technique. Additionally, when evaluating surgical techniques, future studies should assess maxillary growth, as this also may be affected by surgical techniques.

One of the most important findings of the present thesis was the influence of cleft width on various outcomes. This finding should be explored further in future research. Moreover, it is essential to verify the association between cleft width and PCC and PCC-A seen in Study IV.

Articulation ability is probably not solely dependent on cleft type or width. Other factors, such as meta-phonological ability and language ability, may have an impact and should be considered in future studies.

Given the large proportion of children displaying articulation difficulties, which are, in some cases, very severe difficulties, more intervention studies are warranted. We need to identify effective and tolerable methods in relation to the burden for patients and their families.

Conclusions

The present thesis demonstrates various factors that can influence the rate of secondary palatal surgery and speech outcomes.

- Cleft width has a significant impact on the rate of secondary palatal surgery, velopharyngeal function, and articulation proficiency.
- Adopted children seem to be at a higher risk of more persistent articulation errors, still present at ten years of age. While the reasons for this are complex and multifactorial, one of the contributing factors may be the timing of their palatoplasty.
- Developmental articulation errors are common in children with cleft lip and/or palate, regardless of cleft type or whether they were adopted, and intervention should thus target both developmental and cleft-related articulation errors.
- Orderly implementation with structured training when introducing new surgical techniques is crucial.
- It was more common for children with bilateral cleft lip and palate to have articulation difficulties, but there was no difference between cleft types regarding the type of errors made.

Sammanfattning på svenska

Barn som föds med gomspalt behöver genomgå gomkirurgi för att ha möjlighet att utveckla ett normalt tal. I Sverige genomförs gomkirurgi i antingen ett eller två steg. Vid enstegsförfarande sluts hela gommen mellan 9–15 månaders ålder och vid tvåstegsförfarande sluts mjuka gommen vid sex månaders ålder och hårda gommen vid två års ålder. Omhändertagandet är sedan multiprofessionellt och pågår upp i vuxen ålder. Trots tidigt kirurgiskt omhändertagande kan sekundära gomoperationer bli aktuella. Gomfunktionen kan, trots kirurgi, vara otillräcklig och barnet behöver då genomgå så kallad talförbättrande kirurgi. Ibland uppstår läkningsproblem efter den primära gomkirurgin och sekundära operationer kan vara aktuella. Till det kommer att ljud- och talutvecklingen ofrånkomligen påverkas av spalten och barn med gomspalt uppvisar mer artikulationsavvikelser än barn utan gomspalt.

Fokus i detta projekt har varit att identifiera olika faktorer som kan påverka behovet av sekundär gomkirurgi och hur talet utvecklas hos barn födda med gomspalt. Flera faktorer har föreslagits påverka utfallet, däribland kirurgisk teknik, ålder vid operation, vilken typ av spalt barnet har samt gomspaltens bredd. Resultaten från olika studier är ibland motsägelsefulla och har inte utforskats tillräckligt.

Studie I studerade effekten av spalttyp och spaltbredd på frekvensen av sekundär gomkirurgi och talvariabler kopplade till gomfunktionen från åldrarna 3 till 16 år. Spaltbredd var förknippad med en ökad frekvens av sekundär gomkirurgi och sämre tal vid tre och fem års ålder. Spalttypen var inte förknippad med frekvensen av sekundär kirurgi eller talutfall vid någon ålder. Det var tydligt att en god gomfunktion vid tre eller fem års ålder inte alltid innebar en god gomfunktion i tonåren och en liten andel som tidigare bedömts ha en god gomfunktion bedömdes vara i behov av en talförbättrande kirurgi vid bedömning vid 16 års ålder.

Studie II undersökte utvecklingen av gomfunktion och artikulation mellan fem och tio års ålder hos barn adopterade från Kina jämfört med svenskfödda barn födda med läpp- käk- och gomspalt. De adopterade barnen genomgick gomplastik betydligt senare än de icke-adopterade. Vid både fem och tio års ålder noterades att de adopterade hade mer och allvarigare artikulationssvårigheter

vilket tyder på ett behov av mer omfattande insatser. Antalet barn i de båda grupperna som bedömdes ha inkompetent gomfunktion var jämförbart, dock var det färre av de adopterade barnen som bedömdes ha en fullgod gomfunktion.

Studie III utforskade olika typer av kirurgiska tekniker för slutning av den mjuka gommen och dess effekt på artikulation, velofarynxfunktion och behov av sekundär gomkirurgi hos femåringar födda med unilateral läpp-, käk-, och gomspalt. Hur spaltbredd och kirurgens erfarenhet påverkade talet och behov av sekundär gomkirurgi undersöktes också. Resultaten visade att spaltbredd men inte kirurgisk teknik var signifikant associerad till frekvensen av sekundär gomkirurgi och gomfunktion men var inte associerat med artikulationsförmågan. En av fyra kirurger var verksamma över hela studieperioden och dennes resultat förbättrades över tid vilket kan förklaras med ökad erfarenhet. De förbättrade resultaten skulle dock även kunna förklaras med att en ny teknik infördes och att det, möjligen i kombination med ökad erfarenhet, gav lägre frekvens av sekundär gomkirurgi och bättre gomfunktion.

Studie IV undersökte sambandet mellan spalttyp och spaltbredd med frekvensen av sekundär gomkirurgi och artikulation hos femåringar födda med uni- eller bilateral läpp-, käk-, och gomspalt eller spalt omfattande både mjuka och hårda gommen. Spaltbredden men inte spalttypen var signifikant associerat med artikulationsförmåga där bredare spalter hade lägre andel korrekt artikulerade konsonanter. Varken spalttyp eller bredd var signifikant associerad med frekvensen av sekundär gomkirurgi eller gomfunktion.

Denna avhandling belyser vikten av att ta hänsyn till olika faktorer när man vill predicera frekvensen av sekundär gomkirurgi, gomfunktion och artikulationsförmåga. Spaltbredden var mer avgörande än både kirurgisk teknik och spalttyp. Adopterade barn verkar löpa en högre risk för mer ihållande artikulationsfel, vilket, åtminstone delvis, kan bero på deras senare gomslutning. Vid fem års ålder uppvisar samtliga grupper både spaltrelaterade och utvecklingsrelaterade talavvikelser och båda avvikelserna behöver beaktas när talbehandling genomförs.

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Paper I



Greater Palatal Cleft Width Predicts an Increased Risk for Unfavorable Outcomes in Cleft Palate Repair

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Abstract

Objective: To investigate the impact of cleft width and cleft type on the need for secondary surgery and velopharyngeal competence from a longitudinal perspective.

Design: Retrospective, longitudinal study.

Setting: A single multidisciplinary craniofacial team at a university hospital.

Patients: Consecutive patients with unilateral or bilateral cleft lip and palate and cleft palate only ($n = 313$) born from 1984 to 2002, treated with 2-stage palatal surgery, were reviewed. A total of 213 patients were included.

Main Outcome Measures: The impact of initial cleft width and cleft type on secondary surgery. Assessment of hypernasality, audible nasal emission, and glottal articulation from routine follow-ups from 3 to 16 years of age. The assessments were compared with reassessments of 10% of the recordings.

Results: Cleft width, but not cleft type, predicted the need for secondary surgery, either due to palatal dehiscence or velopharyngeal insufficiency. The distribution of cleft width between the scale steps on a 4-point scale for hypernasality and audible nasal emission differed significantly at 5 years of age but not at any other age. Presence of glottal articulation differed significantly at 3 and 5 years of age. No differences between cleft types were seen at any age for any speech variable.

Conclusions: Cleft width emerged as a predictor of the need for secondary surgery as well as more deviance in speech variables related to velopharyngeal competence during the preschool years. Cleft type was not related to the need for secondary surgery nor speech outcome at any age.

Keywords

cleft width, cleft type, velopharyngeal competence, secondary surgery, velopharyngeal function, speech production, nasality

Introduction

Velopharyngeal competence (VPC) is one of the most important outcome measures after cleft palate repair and is crucial for developing good speech. Several factors impact VPC or velopharyngeal insufficiency (VPI), but predictors for outcome measures have only been partially clarified. It has been proposed that both cleft width and cleft type predict various outcome measures (Sullivan et al., 2009; Mahoney et al., 2013; Choa et al., 2014; Ha et al., 2015; Yuan et al., 2016; Wu et al., 2017).

The majority of studies investigating the impact of cleft width have focused on the relation to surgical outcomes, such as fistula rate or frequency of secondary speech-improving surgery. Increased fistula rates have been reported with cleft

widths greater than 15 mm (Parwaz et al., 2009) and 13 mm, respectively (Landheer et al., 2010). Increased cleft width was found to increase the need for speech-improving surgery (Mahoney et al., 2013; Yuan et al., 2016). Cleft width was the

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independent predictor for speech-improving surgery with a higher frequency in patients with unilateral cleft lip and palate (UCLP) and bilateral cleft lip and palate (BCLP) than in those with cleft palate only (CP) (Mahoney et al., 2013). Botticelli and colleagues (2020) used the actual assessment of VPC as the outcome measurement. They found a positive correlation between cleft width at birth in patients with UCLP and degree of VPI at 5 years of age (Botticelli et al., 2020).

Other studies have investigated the relationship between cleft type and need for speech-improving surgery due to VPI. Severe cleft types were associated with an increase of cleft specific characteristics (Choa et al., 2014). Increased nasality in patients with more severe cleft type was also seen, but it was not a statistically significant association. A higher incidence of speech-improving surgery was seen in more severe clefts according to the Veau classification (Sullivan et al., 2009). However, another study demonstrated slightly worse results in patients with less severe clefts according to Veau classification (Kirschner et al., 1999). A comparison between children with UCLP and BCLP showed no significant differences regarding hypernasality or audible nasal emission (Van Lierde et al., 2002).

Most studies are carried out during the preschool years, and the outcomes in a longitudinal perspective remain to be examined. Also, studies based on standardized speech assessment are scarce, and therefore the knowledge of how cleft width and cleft type correspond to the actual speech outcome is still to be explored. This study aimed to investigate the impact of both cleft width and cleft type on the need for secondary surgery and speech variables related to VPC in a longitudinal perspective.

Methods

The Regional Ethics Committee in Uppsala approved this study (Reference no.: 2017/457).

Participants

Patients treated for cleft lip and/or palate at Uppsala University Hospital were retrospectively studied. The study included patients born from 1984 to 2002, treated according to a uniform protocol with respect to both surgical technique and timing for 2-staged palatal surgery, and followed up to 16 years of age. The inclusion criteria were UCLP, BCLP, or CP, treated with soft palate reconstruction at 6 months of age and closure of the residual cleft at 2 years of age. Syndromic clefts and patients with Pierre Robin sequence were left included.

A consecutive series of 313 patients meeting the diagnostic criteria were retrieved from administrative databases. Exclusion criteria were: (1) missing data, that is, missing medical records or dental cast ($n = 27$); (2) transfer of care to another cleft center ($n = 12$); (3) not treated according to the original surgical protocol ($n = 9$); (4) operated with one-stage palatoplasty ($n = 11$); (5) cleft in the soft palate only, that is, only requiring one primary operation ($n = 20$); (6) cleft in lip and palate but not in the primary palate ($n = 4$); (7) inadequate speech for reliable assessment of VPC ($n = 7$); and (8) death ($n = 10$).

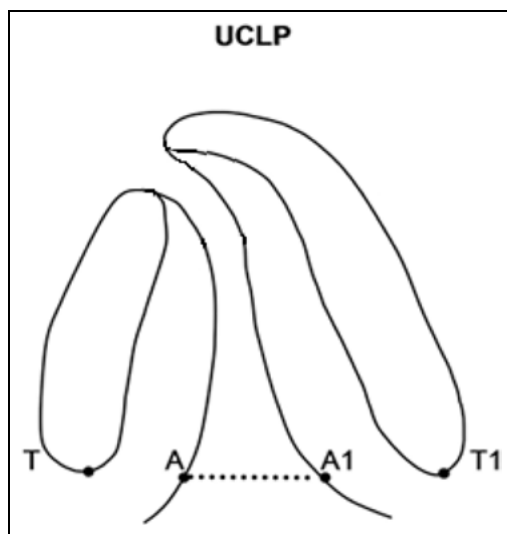


Figure 1. Schematic drawing of an infant maxillary dental cast with UCLP. T-T1, maxillary arch width, measured as the transverse distance between the tuberosity points on each side; A-A1, width of the cleft at the level of T-T1; UCLP, unilateral cleft lip and palate.

Surgery

The surgical protocol at Uppsala University Hospital has since 1984 included a 2-stage palatal reconstruction, with primary soft palatoplasty at 6 months of age and closure of the residual cleft at age 2 years. Obturators were not used. During the period of this study, the reconstruction of the soft palate entailed releasing the nasal layer and palatal muscles in one piece from the posterior part of the hard palate and straight 2-layer closure in the midline. The residual cleft was closed in 2 layers, and relaxing incisions inside the dentition were used if needed. Pharyngeal flap was practiced for secondary speech improvement surgery. Ninety-eight percent of the surgeries were performed by surgeons trained and anchored in the cleft palate team and thus calibrated in surgical technique and execution.

Cleft Width Measurement

Cleft width was measured on dental study casts obtained in connection with the first surgical procedure, either the primary lip repair or primary palatoplasty. The reference points and linear measurements used in this study have been described in previous publications (Hellquist & Skoog, 1976; Friede et al., 1993; Reiser et al., 2010). T-T1 represents the maxillary arch width, measured as the transverse distance between the tuberosity points on each side. A-A1 represents the width of the cleft at the level of T-T1; see Figure 1. The cleft width was calculated as the ratio $A-A1/T-T1$. Distances were measured to the nearest 0.01 mm using a digital caliper. The same operator

(FJ) performed all measurements. Fifteen percent of the dental casts were randomly chosen for duplicated measurement to calculate intrarater agreement.

Speech Assessment at Routine Follow-Up

Speech assessments are performed at 3, 5, 10, and 16 years of age at the CLP-team routine follow-ups by a speech and language pathologist (SLP) specialized in cleft palate speech. Over the years, a variety of speech materials has been used. The main difference between the speech materials was the composition of sounds in words, and words in sentences are, for example, “pappa tittar på TV” in the earlier version compared to “Titti tittar på TV” in the current version. In common for all versions are that they all consist of words and sentences with high-pressure consonants. The speech material currently used consists of 59 words and 8 sentences with high-pressure consonants, which is a bit more extensive than previous materials, which consisted of approximately 20 to 30 words and 4 to 5 sentences. The speech is recorded using an audio recorder (a portable tape recorder or Zoom H4n) or a PC with Soundswell software (Saven Hitech), in combination with a condenser microphone (Sennheiser MKE 2 P-C, Philips speechmike 6264 or Rode NT4). The assessments are based on these audio recordings, graded, and documented in the patient chart.

Hypernasality and audible nasal emission were rated on a 5-point ordinal scale which was then changed to a 4-point ordinal scale in 2016. Glottal articulation was also rated on the 5-point ordinal scale until the year 2000. Since then, glottal articulation has been transcribed and reported in number of occurrences.

Speech Analysis and Reassessment of Speech

Documented assessments of hypernasality, audible nasal emission, and glottal articulation were analyzed. Glottal articulation was dichotomized into present or absent for the purposes of this study. It was considered absent if rated with the 2 lowest scale steps (0 or 1) on the 5-point scale or transcribed with a maximum of 2 occurrences. The scale steps 0 and 1 from the 5-point scale were pooled into scale step 0 in the 4-point scale. Results will be presented according to the 4-point scale.

For assessment of reliability, data from the medical records were compared with reassessments of recordings. Audio recordings from 10% in each group of cleft type were randomly chosen for reassessment. The audio recordings were edited and transferred to .wav files in Praat (Boersma & Weenink, 2018). Personal data were deleted from the sound files to ensure anonymity. The files were coded and randomized.

An external SLP (EH) with long experience of assessing cleft-related speech performed the reassessment. The recordings were listened to through headphones and as many times needed. Hypernasality and audible nasal emission were rated on the 4-point ordinal scale used from 2016. Glottal articulation was noted as present if 3 or more occurrences were noted. Recordings of 30% of the randomly chosen sound files were duplicated for assessment of intrarater agreement.

Statistical Analysis

Logistic regression analysis was used to investigate the impact of cleft width and cleft type on secondary surgery. The distribution of cleft width between scale steps was analyzed with Kruskal-Wallis test. For comparison between the cleft types, chi-square test was used. Agreement between speech ratings from the medical records and the external listener was calculated using single measures intraclass correlation coefficient (ICC) with a 2-way mixed-effects model. The ICC was also used to calculate intrajudge agreement for both speech and cleft width. According to Cicchetti, the strength of agreement was interpreted as <.40 is poor, .40 to .59 is fair, .60 to .74 is good, and .75 to 1.00 is excellent (Cicchetti, 2001). Individuals who had secondary surgery due to fistulas or dehiscence were included in the speech analyses, while individuals who had had speech-improving surgery at the time of the routine control at any age were excluded from the analyses of speech variables at that age and later. Statistical analysis was performed in IBM SPSS Statistics, version 23. The level of significance was determined to be $\alpha < 0.05$.

Agreement Between Medical Records and the External SLP

The single measures ICC calculated absolute agreement between assessments from medical records and the external SLP. The agreement for hypernasality and audible nasal emission was fair (0.51, 95% CI: 0.31-0.67 and 0.51, 95% CI: 0.30-0.67). The agreement for glottal articulation was 100%.

Intrarater Agreement

Intrarater agreement for hypernasality and audible nasal emission was excellent (0.96, 95% CI: 0.90-0.99 and 0.97, 95% CI: 0.90-0.99). The intrarater agreement was 100% for glottal articulation.

Intrarater agreement for measuring cleft width was excellent (0.96, 95% CI: 0.92-0.98).

Results

Two hundred thirteen individuals met the inclusion criteria, with a male to female ratio of 1.5:1, 86 with UCLP (58 boys, 28 girls), 37 with BCLP (31 boys, 6 girls), and 90 with CP (38 boys, 52 girls). Nine patients were diagnosed with a syndrome, and 15 had Pierre Robins sequence.

Soft palate closure was performed at a mean age of 7.2 months (median: 6 months, range: 4 to 17 months, standard deviation: [SD] 2.2 months) and hard palate closure at a mean age of 24.9 months (median: 24 months, range: 9 to 45 months, SD: 3.9 months).

In total, 23 (11%) patients required secondary palatoplasty due to partial or complete dehiscence of the soft palate (10 with UCLP, 3 with BCLP, 10 with CP). In 9 of these patients, a pharyngeal flap had to be performed at the salvage palatoplasty, and thus not primarily as a speech-improving surgery. Another

Table 1. Characteristics of Participants.

Parameter	Cohort, n = 213 (included in speech analyses, n = 140)					
	UCLP		BCLP		CP	
	Surgery analyses n (%)	Speech analyses n (%)	Surgery analyses n (%)	Speech analyses n (%)	Surgery analyses n (%)	Speech analyses n (%)
Sex						
Male	58 (67)	37 (61)	31 (84)	18 (90)	38 (42)	25 (42)
Female	28 (33)	24 (39)	6 (16)	2 (10)	52 (58)	34 (58)
Rupture of soft palate						
No	76 (88)	55 (90)	34 (92)	17 (85)	80 (89)	51 (86)
Yes	10 (12)	6 (10)	3 (8)	3 (15)	10 (11)	8 (14)
Fistula						
No	77 (90)	53 (87)	32 (86)	19 (95)	76 (84)	51 (86)
Yes	9 (10)	8 (13)	5 (14)	1 (5)	14 (16)	8 (14)
Speech improving surgery						
No	64 (74)	48 (79)	27 (73)	16 (80)	72 (80)	47 (80)
Yes	22 (26)	13 (21)	10 (27)	4 (20)	18 (20)	12 (20)

Abbreviations: BCLP, bilateral cleft lip and palate; CP, cleft palate only; UCLP, unilateral cleft lip and palate.

9 of the 23 patients with dehiscence required a pharyngeal flap due to VPI later on. Twenty-eight (13%) patients underwent fistula repair (9 with UCLP, 5 with BCLP, 14 with CP). In total, 50 (23%) patients required speech-improving surgery at a mean age of 7 years and 6 months (median: 6 years and 3 months, range: 3 years and 4 months-15 years and 3 months, SD: 3 years and 1 month). Forty-six patients received a pharyngeal flap (21 with UCLP, 10 with BCLP, 15 with CP), and 4 received rerepair of the soft palate (3 with CP, 1 with UCLP). Two of the patients with a rerepair required a pharyngeal flap later on (Table 1). Nasendoscopy was performed in all patients where the perceptual speech indicated a need for speech-improving surgery.

All 213 patients were included in analyzing the need for secondary surgery due to palatal dehiscence or VPI. One hundred forty (66%) of those had at least 3 of 4 possible speech assessments and were included in the speech outcome analysis (61 with UCLP, 20 with BCLP, 59 with CP). Seventy-three had fewer than 3 assessments due to difficulty participating, technical failure during recording, or that assessment was not made on time (3 years, ±3 months; 5 years, ±6 months; 10 ± 1 year; and 16 years, ± 1 year). In total, 492 assessments were made in the 140 patients with 3 or 4 assessments.

Logistic regression analysis demonstrated that greater cleft width was significantly associated with need of secondary surgery due to dehiscence (odds ratio [OR] 1.102, 95% CI: 1.041-1.167, *P* < .001) but cleft type was not (UCLP vs BCLP: OR .373, 95% CI: .088-1.585, *P* = .182; UCLP vs CP: OR 1.733, 95% CI: .625-4800, *P* = .290), see Table 2A. Mean cleft width in those in need of secondary surgery was 28% (range 10%-44%). In those not needing secondary surgery, mean cleft width was 23% (range 3%-57%).

After excluding those who had secondary surgery due to dehiscence, 194 individuals were included in the logistic regression analysis showing that greater cleft width was significantly associated with need of speech-improving surgery

Table 2A. Logistic Regression Analysis to Estimate the Association Between (a) Cleft Width and (b) Cleft Type and the Need for Secondary Surgery Due to Dehiscence of Soft Palate.

Variable	<i>P</i>	Odds ratio	95% CI
Cleft width	<.001	1.10	1.04-1.17
UCLP	.15		
BCLP	.18	.37	.09-1.59
CP	.29	1.73	.63-4.80
Constant	<.001	.009	

Abbreviations: BCLP, bilateral cleft lip and palate; CP, cleft palate only; UCLP, unilateral cleft lip and palate.

Table 2B. Logistic Regression Analysis to Estimate the Association Between (a) Cleft Width and (b) Cleft Type and the Need for Speech Improving Surgery.

Variable	<i>P</i>	Odds ratio	95% CI
Cleft width	.017	1.06	1.01-1.11
UCLP	.92		
BCLP	.75	.85	.32-2.29
CP	.87	1.07	.47-2.48
Constant	<.001	.08	

Abbreviations: BCLP, bilateral cleft lip and palate; CP, cleft palate only; UCLP, unilateral cleft lip and palate.

(OR 1.058, 95% CI: 1.010-1.107, *P* = .017) but cleft type was not (UCLP vs BCLP: OR .854, 95% CI: .320-2.278, *P* = .752; UCLP vs CP: OR 1.074, 95% CI: .466-2.479, *P* = .867), see Table 2B. Mean cleft width in those in need of speech-improving surgery was 26% (range 8%-57%). In those not needing speech-improving surgery, mean cleft width was 22% (range 3%-42%).

An analysis with Kruskal-Wallis test revealed a significant difference between the distribution of cleft width between the

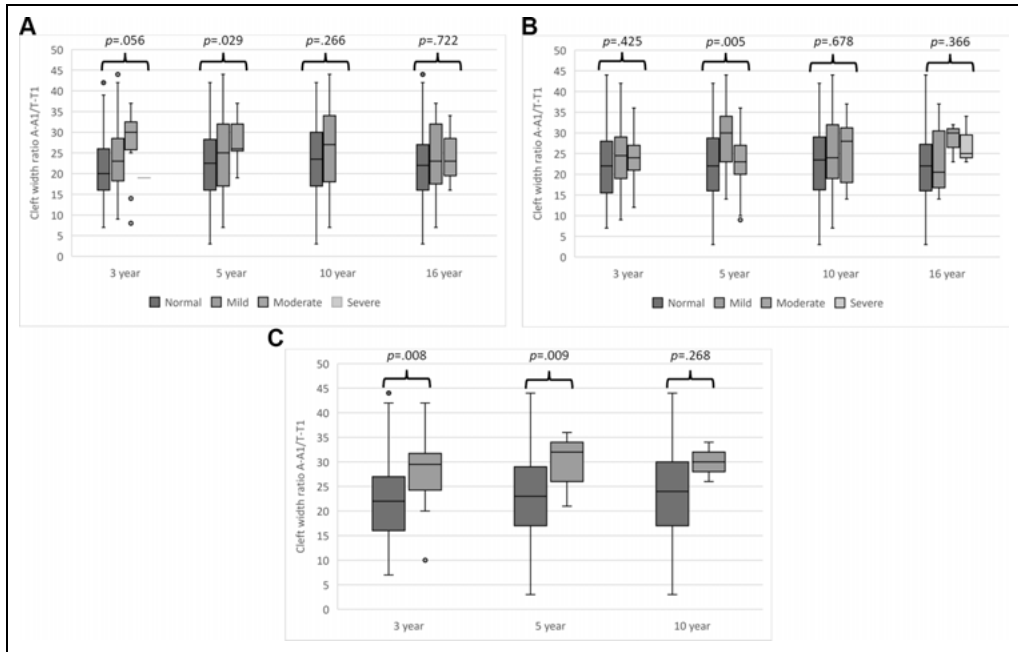


Figure 2. A, Median cleft width distributed on the scale steps for hypernasality at the respective follow-up time points. B, Median cleft width distributed on the scale steps for nasal emission at the respective follow-up time points. C, Median cleft width distributed between presence and absence of glottal articulation at the respective follow-up time points. No one had glottal articulation at 16 years of age.

scale steps for hypernasality at 5 years of age, but not at any other age (3 years: $H[3] = 7.55, P = .056$; 5 years: $H[2] = 7.08, P = .029$; 10 years: $H[1] = 1.24, P = .266$; 16 years: $H[2] = 0.65, P = .722$). Similarly, audible nasal emission differed significantly at 5 years of age, but not at any other age (3 years: $H[2] = 1.71, P = .425$; 5 years: $H[2] = 10.76, P = .005$; 10 years: $H[2] = 0.78, P = .678$; 16 years: $H[3] = 3.17, P = .366$). Glottal articulation differed significantly at 3 and 5 years of age but not at 10 years of age (3 years: $H[1] = 6.95, P = .008$; 5 years: $H[1] = 6.77, P = .009$; 10 years: $H[1] = 1.23, P = .268$). No individual had glottal articulation at 16 years of age, see Figure 2A-C. Taken together, wider cleft width was related to inferior speech outcomes at 5 years of age.

Chi-square calculation showed no significant differences between the cleft types at any age for hypernasality (3 years: $\chi^2_6 = 3.16, P = .788$; 5 years: $\chi^2_4 = .42, P = .981$; 10 years: $\chi^2_2 = .26, P = .876$; 16 years: $\chi^2_4 = 6.02, P = .197$), audible nasal emission (3 years: $\chi^2_4 = .31, P = .989$; 5 years: $\chi^2_4 = 7.75, P = .101$; 10 years: $\chi^2_4 = 2.54, P = .637$; 16 years: $\chi^2_6 = 7.18, P = .305$), or glottal articulation (3 years: $\chi^2_2 = 3.11, P = .212$; 5 years: $\chi^2_2 = 2.93, P = .231$; 10 years: $\chi^2_2 = 2.51, P = .286$; 16 years: no glottal articulation was noted), see Figure 3A-C.

Discussion

This study investigated the impact of cleft width and cleft type on the need for secondary surgery and speech outcome in a longitudinal perspective. An increasing need for secondary surgery, both due to partial or complete dehiscence and VPI, was associated with increasing cleft width but not with cleft type. The results are consistent with previous studies (Mahoney et al., 2013; Yuan et al., 2016), indicating that cleft width rather than cleft type predicts unfavorable outcomes. Moreover, studies investigating the effect of cleft type on outcomes in cleft palate repair, without considering cleft dimensions, have shown contradictory results (Kirschner et al., 1999; Van Lierde et al., 2002; Sullivan et al., 2009; Choa et al., 2014), which also implies that cleft width is a stronger predictor for surgical outcomes compared to cleft type. Comparing the results from different studies is complicated by variability in surgical methods, outcome parameters, inter-rater assessments, and statistical analyses. However, the relation of cleft width with outcomes identified in this study seems to be in line with a compiled analysis of previous research.

In this study, cleft type did not affect the outcome of speech variables related to VPC. The final outcome of provided cleft care with respect to speech was not related to initial cleft width.

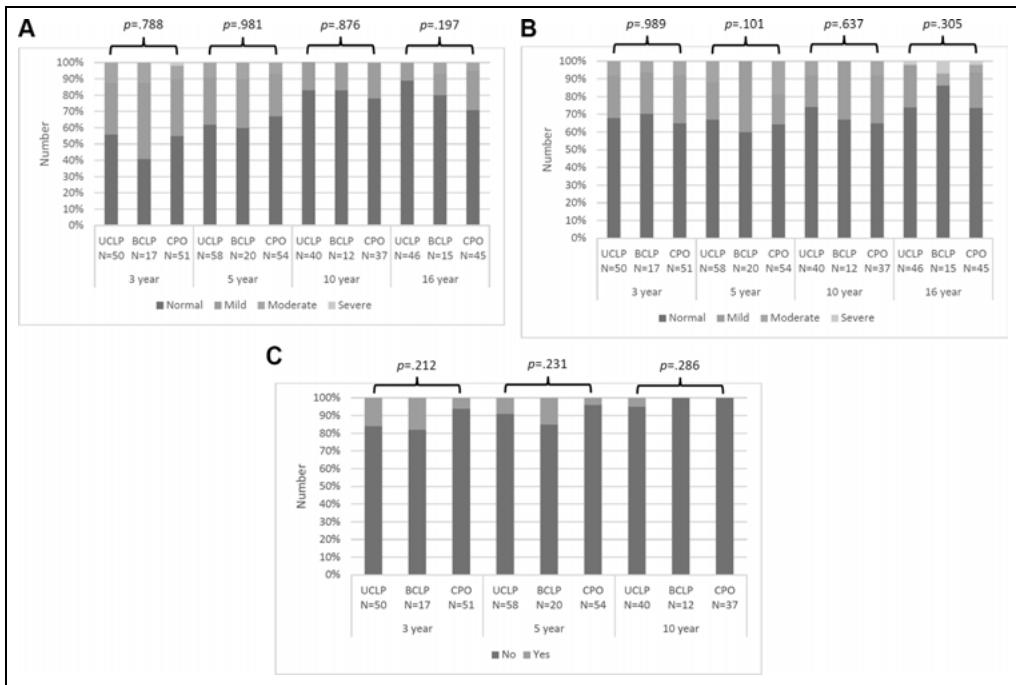


Figure 3. A, Rating of hypernasality at different follow-up time points divided in cleft type. B, Rating of nasal leakage at different follow-up time points divided in cleft type. C, Presence of glottal articulation at different follow-up time points divided in cleft type. No one had glottal articulation at 16 years of age.

However, patients with wider clefts needed significantly more speech-improving surgery. At 3 and 5 years of age, increasing cleft width was associated with more severe deviance in all speech variables. Hypernasality increased significantly with increasing cleft width at 5 years of age. A clear trend of increasing hypernasality in wider clefts, although not statistically significant, was also noticed at 3 years of age. At both 3 and 5 years of age, glottal articulation was significantly associated with increasing cleft width. Audible nasal emission was not related to cleft width at 3 years of age, which is most probably explained by the increased glottal articulation in patients with wider clefts. At 5 years of age, cleft width was greater only in patients with mild audible nasal emission. Audible nasal emission can also be a consequence of a fistula or the remaining cleft in the alveolar ridge and, therefore, not necessarily a sign of actual VPI. Sufficient VPC at 5 years of age did not equal sufficient VPC throughout the years. Pertaining to this, 34% of the patients receiving speech-improving surgery had their operation after 7 years of age. Furthermore, 6 individuals were rated with moderate or severe degree of hypernasality and/or audible nasal emission at 16 years of age and were all recommended speech-improving surgery. Half of them had shown no signs of VPI, and the other half had shown only minor signs of VPI at

the earlier routine controls. Thus, in a proportion of patients, a need for speech-improving surgery developed over the years, underlining the importance of routine controls right up to adulthood.

This study's major strength is that the effect of cleft width and cleft type on speech variables related to VPC is evaluated longitudinally and compared at fixed ages. Many previous studies have been hampered by too short and variable follow-up, as well as an inadequately described methodology for speech assessment. Moreover, there is a lack of longitudinal data based solely on each patient's repetitive assessments, which is vital to analyze the dynamics of speech development in patients with cleft over time. Another important strength is the relatively large cohort treated according to a uniform surgical protocol, eliminating the variable of surgical methodology and reducing the risk for negative findings being caused by inadequate power.

Reliable assessments of speech variables include high-quality audio recordings with standardized speech material. Preferably, at least 2 independent and blinded raters should perform a reassessment of audio recordings to enable inter- and intrareliability analysis (Sell, 2005). Because of variable technical quality and different length of speech samples of saved

audio recordings, reassessment was not possible for the whole material. This limitation was addressed by performing reassessments in 10% of the available recordings with adequate quality and number elicitations of words and/or sentences. The interrater reliability was fair for both hypernasality and audible nasal emission, and the speech data should, accordingly, be interpreted with some caution. Hypernasality is known to be a difficult variable to reach high interrater reliability (Watterson et al., 2007), and studies have shown varying results (Lewis et al., 2003; Brunnegård & Lohmander, 2007; Brunnegård et al., 2009; Lohmander et al., 2012; Lohmander et al., 2017; Yamashita et al., 2018). Two previous studies presented somewhat higher inter-rater reliability for audible nasal emission than the present study (Persson et al., 2006; Brunnegård & Lohmander, 2007).

Other limitations of this study are the lack of data on other potential confounders such as hearing status and data on syndromes. Reduced hearing may impede speech development, and this could theoretically interfere in the analysis of effects of cleft width and type on speech. We found a relatively low percentage of patients diagnosed with a syndrome, which most probably relates to uncertain syndrome diagnosis during the earlier years of the study period. Therefore, we chose to include patients with a verified syndrome diagnosis or with Pierre Robin sequence. Patients where speech assessment was not possible due to inadequate speech development were excluded from the analysis, signifying that syndromic patients with markedly delayed development were, in effect, excluded. The increased frequency of speech-improving surgery and the speech deviance during preschool years are coherent and indicate that cleft width is an independent predictor for speech outcomes.

Conclusion

Cleft width emerged as a predictor for secondary surgery, both for dehiscence and VPI, as well as for speech variables related to VPC at 5 years of age. Cleft type was not significantly associated with the need for secondary surgery, and speech variables did not differ at any age between the cleft types. This indicates that cleft width, rather than cleft type, is a more reliable predictor. Sufficient VPC at 5 years of age did not equal sufficient VPC throughout the years, and a proportion of patients developed a need for speech-improving surgery over the years, underlining the importance of routine controls right up to adulthood.

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Paper II



RESEARCH REPORT

Longitudinal data on speech outcomes in internationally adopted children compared with non-adopted children with cleft lip and palate

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Abstract

Background: At the beginning of the 21st century, international adoptions of children with cleft lip and/or palate increased dramatically in Sweden. Many children arrived partially or totally unoperated, despite being at an age when palatoplasty has usually been performed. To date, the speech development of internationally adopted (IA) children has been described up to age 7–8 years, but later development remains unstudied.

Aims: To investigate speech development between ages 5 and 10 years in children born with cleft lip and palate (CLP) adopted from China and to compare them with non-adopted (NA) children with CLP. A secondary aim was to compare the frequencies of secondary palatal surgery and number of visits to a speech and language pathologist (SLP) between the groups.

Methods & Procedures: In a longitudinal study, 23 IA children from China were included and matched with 23 NA children born in Sweden. Experienced SLPs blindly reassessed audio recordings from routine follow-ups at ages 5 and 10 years. Velopharyngeal function (VPF) was assessed with the composite score for velopharyngeal competence (VPC-Sum) for single words and rated on a three-point scale (VPC-Rate) in sentence repetition. Target sounds in words and sentences were phonetically transcribed. Per cent correct consonants (PCC) were calculated at word and sentence levels. For in-depth analyses, articulation errors were divided into cleft speech characteristics (CSCs), developmental speech characteristics (DSCs) and s-errors. Information on secondary palatal surgery and number of visits to an SLP was collected.

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Outcomes & Results: VPF differed significantly between the groups at both ages when assessed with VPC-Sum, but not with VPC-Rate. Regardless of the method for assessing VPF, a similar proportion in both groups had incompetent VPF but fewer IA than NA children had competent VPF at both ages. IA children had lower PCC at both ages at both word and sentence levels. More IA children had CSCs, DSCs and s-errors at age 5 years, and CSCs and s-errors at age 10. The development of PCC was significant in both groups between ages 5 and 10 years. The proportion of children receiving secondary palatal surgery did not differ significantly between the groups, nor did number of SLP visits.

Conclusions & Implications: CSCs were more persistent in IA children than in NA children at age 10 years. Interventions should target both cleft and DSCs, be comprehensive and continue past the pre-school years.

KEYWORDS

articulation, international adoption, longitudinal, velopharyngeal function

WHAT THIS PAPER ADDS

What is already known on this subject

- At the beginning of the 21st century, IA children with cleft lip and/or palate arrived in Sweden partially or totally unoperated, despite being at an age when palatoplasty has usually been performed. Studies up to age 7–8 years show that adopted children, compared with NA peers, have poorer articulation skills, demonstrate both cleft-related and developmental articulation errors, and are more likely to have velopharyngeal incompetence. Several studies also report that adopted children more often require secondary palatal surgery due to fistulas, dehiscence or velopharyngeal incompetence compared with NA peers.

What this paper adds to existing knowledge

- This longitudinal study provides additional knowledge based on longer follow-ups than previous studies. It shows that the proportion of children assessed to have incompetent VPF was similar among IA and NA children. It was no significant difference between the groups regarding the proportion that received secondary palatal surgery. However, fewer IA children were assessed to have a competent VPF. Developmental articulation errors have ceased in most IA and all NA children at age 10 years, but significantly more adopted children than NA children still have cleft-related articulation errors.

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

- Speech and language therapy should target both cleft-related and developmental articulation errors. When needed, treatment must be initiated early, comprehensive, and continued past the pre-school years, not least for adopted children.

INTRODUCTION

At the beginning of the 21st century, the number of internationally adopted (IA) children with special needs, including those born with cleft lip and/or palate (CLP), increased dramatically in, for example, Sweden and the United States, with the vast majority originating from China (Goldstein et al., 2014; Hansson et al., 2012; Kaye et al., 2019; Shay et al., 2016; Sullivan et al., 2014; Swanson et al., 2014). The incidence of CLP in Sweden is 2/1000 live births (Hagberg et al., 1998), which means approximately 150–200 children per year. In 2007, 24 children with CLP were adopted to Sweden. In 2009 the number had increased to 98 children, representing 14% of the total number of adopted children 2009 (Stållhand, 2010). Mean age at arrival between 20 and 30.5 months has been reported (Goldstein et al., 2014; Kaye et al., 2019; Larsson et al., 2017; Swanson et al., 2014). The cleft teams met new challenges as many of the IA children arrived partially or totally unoperated despite being at an age when non-adopted (NA) children would have had their palatoplasty done. Moreover, IA children undergo a language shift and are just settling in with their new families at the time of their first palatoplasty, usually performed within 3 months after arrival (Hansson et al., 2012; Larsson et al., 2020; Shay et al., 2016).

Velopharyngeal insufficiency (VPI) has been associated with increasing age at primary palatoplasty in IA children (Follmar et al., 2015; Pet et al., 2018; Sullivan et al., 2014). It has also been shown in several studies that more IA than NA children need secondary surgery due to fistulas, dehiscence, or VPI (Hansson et al., 2012; Morgan et al., 2018; Swanson et al., 2014), but not always at a significant level (Pet et al., 2018). In addition, IA children with CLP have been described as having poorer language skills (Kaye et al., 2019; Scherer et al., 2018) and articulation (Larsson et al., 2017, 2020; Morgan et al., 2018) compared with NA children with CLP.

Larsson et al. (2017) found that 3-year-old IA children ($n = 14$) with unilateral cleft lip and palate (UCLP) had significantly more articulation errors than their NA peers ($n = 18$). Furthermore, more IA children than NA children were judged to have an incompetent velopharyngeal function (VPF). A comparison between IA and NA children at age 5 years showed that IA children more often than NA children exhibited articulation errors related to the cleft as well as errors related to development, and 52% of IA children had incompetent VPF compared with 25% of NA children (Larsson et al., 2020). Similar results were shown when a comparison between IA and NA children with cleft palate, with or without a cleft lip, was made in three age groups

(3–4, 5–6 and 7–8 years) (Morgan et al., 2018). In all age groups, IA children had poorer articulation skills than NA children. At ages 7–8 years, cleft-related articulation errors were noted in just over half of the IA group compared with just under one-third of the NA US-born children. A larger proportion of the IA children than their NA peers also exhibited developmental phonological errors at ages 7–8 years. Morgan et al. (2018) did not find a significant correlation between age at adoption and articulation outcomes. However, time with sufficient VPF was associated with better articulation skills in both groups. Typically developed NA children without CLP in Sweden score a mean of 95.8% (SD = 4.8, range = 76–100%) correct consonants at word level as 5-year-olds, 97.8% (SD = 3.3, range = 88–100%) as 7-year-olds, and 98.8% (SD = 3.2, range = 86–100%) as 10-year-olds (Lohmander et al., 2017b). A longitudinal study of children with UCLP adopted from China (Larsson et al., 2022) reported a significant improvement between 3 and 7–8 years of age in both per cent consonants correct (PCC) and VPF. Despite the significant improvement reported, 94% of the children had PCC scores > 2 SD below typically developed NA children without CLP at ages 7–8 years. Moreover, developmental speech characteristics (DSCs), to some extent, were still present at ages 7–8 years in 77% of the children.

To date, articulation and VPF have been described and compared between IA and NA children up to age 7–8 years, but what happens after that is still unknown. There is also a lack of knowledge about how articulation and VPF develop over time, as only a few longitudinal studies, including IA children, have been published. The results from previous studies are consistent: IA children have poorer articulation skills and more often have VPI than their NA peers. More studies at later ages are required to understand the full extent of the consequences of the delayed speech development previously described at earlier ages. This study aimed to investigate how VPF and articulation skills develop in a longitudinal perspective between ages 5 and 10 years in children with CLP adopted from China compared with children with CLP born in Sweden. A secondary aim was to compare VPF, articulation, the proportions of secondary palatal surgery, and the number of visits to a speech and language pathologist (SLP) between IA and NA children.

The following research questions were posed:

How do VPF and articulation develop over time?

Are there differences between IA and NA children regarding (1) VPF, (2) articulation, (3) need for secondary palatal surgery or (4) number of visits to an SLP?

METHODS

The Regional Ethics Committee in Uppsala approved this study (Reference no.: 2017/457).

Participants

Information about the participants regarding cleft type, age at adoption, surgery, tympanostomy tubes and number of visits to an SLP was retrieved from their medical records. IA children born in the period 2000–09 and treated for CLP at Uppsala University Hospital were reviewed. Only children originating from China were included, as IA children from other countries were rare during the period of increasing international adoptions to Sweden. This resulted in a larger group of IA children with similar ethnicity, geographical and linguistic origin, and thus rendered a more homogenous group. The other inclusion criteria were unilateral or bilateral CLP. A consecutive series of 39 patients, 25 with UCLP and 15 with bilateral cleft lip and palate (BCLP), met the inclusion criteria. A total of 16 children were excluded due to missing or incomplete audio recordings. Thus, 23 children were included, 17 with UCLP and six with BCLP, six girls and 17 boys. The mean age at adoption was 1;9 (years;months) ($SD = 7$ months, range = 0;11–3;4). The IA children were matched with Swedish-born children (NA) based on cleft type and sex. If an adopted child had surgery in Sweden, he/she was also matched on the surgical technique for closure of the soft palate, as the surgical methods used changed during the study period. The NA group was selected from a consecutive series of 105 NA children with UCLP ($n = 76$) or BCLP ($n = 29$) born in the period 2000–10 and treated at Uppsala University Hospital. A random selection was made to prevent bias if more than one Swedish-born child with a complete audio recording matched an adopted child. One included NA child had Van der Woude syndrome.

A total of 12 IA children (52%) were treated with tympanostomy tubes at a mean age of 2;7 (range = 1;1–3;9) and a mean of 8 months after adoption (range = 2–15 months). A total of 16 NA children (70%) were treated with tympanostomy tubes at a mean age of 1;0 (range = 3–27 months).

Surgical treatment

The standard procedure at Uppsala University Hospital since 1984 has been to close the palate in two stages: soft palate closure (SPC) at age 6–8 months and hard palate

closure (HPC) at age 2 years. Before 2008, SPC was performed with an intra-velar veloplasty reinforced by the palatopharyngeal muscle (Henriksson et al., 2005). Since 2008, intra-velar veloplasty has been performed (Sommerlad, 2003, 2006). Speech-improving surgery was performed with a pharyngeal flap. Before such surgery, the patients were evaluated with a speech assessment, nasoendoscopy and, when possible, nasometry. The order of operations was the same in both groups, but the timing differed. A total of 15 (65%) in the IA group and 11 (48%) in the NA group had their secondary alveolar bone grafting before the assessment at age 10 years. See Table 1 for surgical details.

Speech material

Audio recordings from routine follow-ups at ages 5 and 10 years were used for phonetic transcription, rating of hypernasality and perceived VPF. The audio recordings were made with Zoom H4n or a PC with Soundswell software (Saven Hitech, Stockholm, Sweden) and a condenser microphone (Røde NT4, Sydney, Australia or Philips SpeechMike Classic 6264). Mean ages at recording were 5;2 and 10;1 in the IA group (ranges = 4;11–5;8 and 9;8–10;7) and 5;1 and 10;1 in the NA group (ranges = 4;11–5;5 and 9;7–10;5). The speech material consisted of 59 words and 13 sentences from the Swedish Articulatory and Nasality Test (SVANTE) (Lohmander et al., 2017b). The 59 words and the first eight sentences each include target consonants particularly vulnerable to cleft-related speech difficulties (plosives /p/, /b/, /t/, /d/, /k/ and /g/, and voiceless fricatives /s/, and /f/). The first nine words also include high vowels, (/i:/, /u:/ and /u:/). Three sets of speech material were edited from the recordings: (1) 59 single words, each including one target consonant; (2) a nine-word string (the first nine monosyllabic words edited to a string of words with no pause between them) including high vowels for assessment of hypernasality; and (3) 12 sentences used for phonetic transcription of target consonants and rating of VPF—the 13th consisted only of nasals and was therefore excluded. The first eight sentences, including four target consonants each, were phonetically transcribed. All 12 were included in the rating of VPF. Picture naming or reading from a list was used to elicit the words, and sentences were repeated after the SLP. The audio recordings (.wav files) were edited using Praat (Boersma & Weenink, 2018) to contain only the speech from the child. The audio files were then randomized and assigned codes to ensure blinded assessments. The audio files were listened to through high-quality headphones and could be replayed as many times as needed.

TABLE 1 Primary surgeries

	Internationally adopted	Non-adopted
Surgery before adoption, <i>n</i> (%)	12 (52%) lip 7 (30%) lip + palate	–
Age at SPC, mean (SD, range)	2;0 (7, 1;1–3;9)	0;7 (1, 0;6–0;9)
Age at HPC, mean (SD, range)	2;6 (7, 2;0–4;3)	2;1 (2, 1;10–2;8)
Secondary alveolar bone grafting, mean (SD, range)	9;8 (9, 8;3–10;11)	10;0 (10, 8;10–11;8)

Notes: Mean ages and ranges are given as years;months, SD is given in months. HPC, hard palate closure; SPC, soft palate closure.

Speech assessment

Calibration. Before rating hypernasality and a perceptual rating of velopharyngeal competence (VPC-Rate) and the phonetic transcription of target sounds in words and sentences, two training sessions were performed to calibrate the SLPs. Four SLPs with 6–18 years of experience with patients with cleft palate were involved in the assessments, three in the assessment of hypernasality and VPC-Rate, and two in the phonetic transcriptions. SLPs 1 and 2 were from the same centre the participants were recruited from, and SLPs 3 and 4 were from two other centres in Sweden. SLPs 1–3 spent 1.5 h calibrating their ratings of hypernasality and VPC-Rate. They were instructed first to decide if hypernasality and VPF were within normal limits and, if not, rate the degree of hypernasality as mild, moderate or severe, and VPF marginally incompetent or incompetent. They listened to one file at a time, rated them individually and then compared their ratings. SLPs 1 and 4 spent 4 h listening to and discussing their phonetic transcriptions. They listened to five words or one sentence at a time and then compared their transcriptions. In cases of disagreement in the transcriptions or ratings, the SLPs discussed until reaching consensus.

VPF. Two variables to assess VPF were used: VPC-Sum and VPC-Rate (Lohmander et al., 2017a). VPC-Sum was used to assess perceived VPF in words, and VPC-Rate was used to assess perceived VPF in connected speech. VPC-Sum is a composite score encompassing (1) perceptual ratings of hypernasality rated on a four-point scale (normal, mild, moderate, severe); (2) perceptual signs of VPI from transcriptions (nasal emission and weak pressure consonants); and (3) active non-oral consonant errors from transcriptions. The three variables each generate a score of 0–2. Ratings of hypernasality translate to scores as follows: 0 = 0, 1 = 1, and 2–3 = 2. The number of perceptual signs of VPI and active non-oral errors in transcriptions translates to a score as follows: 0–2 occurrences = 0, 3–5 occurrences = 1, and ≥ 6 occurrences = 2. The three scores were added together to calculate VPC-Sum of between 0 and 6 for each child. A score of 0–1 indicates competent VPF, 2–3 indicates marginally incompetent VPF, 4–6 indicates incompetent VPF, and VPC-Sum is reported

as competent, marginally incompetent and incompetent VPF.

VPC-Rate was rated on a three-point scale (competent, marginally incompetent and incompetent). The SLPs rated hypernasality for all children and then VPF, first at age 5 years and then at age 10 years. If a child consistently used glottal articulation, VPF was rated as incompetent. The score for each child was classified based on the majority decision of the three SLP ratings. In case of total disagreement, the middle value was chosen. All audio files, including 30% for intra-rater agreement, were rated by SLPs 1–3.

Phonetic transcription. SLPs 1 and 4 performed a semi-narrow transcription, in this study defined as a phonetic transcription using the International Phonetic Alphabet (IPA) (2015) and the symbols for nasal escape, velopharyngeal friction, weak articulation, voicing, devoicing and active nasal fricative from the extended IPA (2008) symbols for disordered speech. The target sounds in the 59 words and eight sentences were transcribed for all participants by SLP one. A total of 30% of the audio files in each group at each age were randomly selected for intra-rater agreement. For interrater agreement, 50% of the sound files from ages 5 and 10 years were randomly selected and transcribed by SLP 4.

The transcriptions were used to calculate PCC (Shriberg & Kwiatkowski, 1982)¹, where all articulation errors were given the same weight. A consonant was scored correct if the place and manner of articulation were correct. Signs of VPI were disregarded. PCC was calculated at word and sentence levels. Articulation errors were divided into cleft speech characteristics (CSCs) and DSCs, as described in the Scandcleft project (Willadsen et al., 2017). Three or more occurrences signified an error. CSCs were subdivided into non-oral errors (glottal or pharyngeal plosive or fricative, nasal for unvoiced stop or fricative, and active nasal fricative) and oral errors (retracted/backed to palatal/velar/uvular place of articulation and double articulation). Velar fronting (a velar consonant is replaced with a dental or alveolo-dental consonant), stopping (a fricative is replaced with a plosive), and voicing errors (difficulties differentiating between voiced and voiceless consonants) were considered DSCs. An articulation error

TABLE 2 Intra-rater agreement for hypernasality and rating of velopharyngeal competence at ages 5 and 10 years

		Hypernasality		VPC-Rate	
		5 years	10 years	5 years	10 years
Intra-rater	Rater 1	50%	93%	93%	93%
	Rater 2	79%	64%	93%	93%
	Rater 3	71%	93%	93%	86%

Note: VPC-Rate, rating of velopharyngeal competence.

could be counted as both a CSC and a DSC. The consonant /s/ was scored as correct when realized as /s/. All other oral realizations of /s/ that were still realized as a fricative (θ c ζ ξ) were scored as incorrect and categorized as an s-error. Although they may be categorized as CSCs or DSC, s-errors were reported separately.

Statistical analysis

Non-parametric analyses were used for group comparisons. The chi-square test was used to compare VPC-Sum and VPC-Rate between IA and NA children. Mann-Whitney *U*-tests were used to calculate differences in PCC between the groups. The categorical data related to CSCs and DSCs (three or fewer occurrences), the need for speech-improving surgery and number of visits to an SLP were calculated with the Fisher exact test. Statistical analyses were performed in IBM SPSS Statistics, version 28. The level of significance was set at $\alpha < 0.05$.

Agreement

Interrater agreement for hypernasality and VPC-Rate was calculated as the frequency of (1) agreement between all three raters, (2) agreement between two raters and (3) no agreement. Regarding hypernasality at age 5 years, two out of three raters agreed in 97% of the cases (all raters agreed in 38%), with no agreement in 3% of the cases. Two out of three raters agreed in 98% of the cases at age 10 years (all raters agreed in 48%), with no agreement in 2% of the cases. Regarding VPC-Rate, two out of three raters agreed in all cases at both ages (all raters agreed in 65% at age 5 years and 77% at age 10 years). Intra-rater agreement was measured as agreement point by point for both hypernasality and VPC-Rate. Information about intra-rater agreements is shown in Table 2.

Point-by-point comparisons also calculated the inter- and intra-transcriber agreement for the phonetic transcriptions. The agreement was calculated at word and sentence levels at each age and reported as median values. Comparisons were made for correctly articulated target sounds, symptoms of VPI, and the presence of non-oral articula-

TABLE 3 Differences between phonetic transcriptions considered an agreement

/ʔ bʔ/ (glottal versus glottal reinforcement)
/ʔ ʔ ^a /
/c k/ /j g/

^aNotes: Omission of target consonant.

tion. In calculating inter- and intra-transcriber agreement, minor differences were considered an agreement (Table 3). Table 4 provides detailed information about inter- and intra-transcriber agreements.

RESULTS

VPF

VPC-Sum. There was a significant difference between the groups at age 5 years ($p = 0.031$). At age 10 years, the difference between the groups remained significant ($p = 0.003$). Excluding children with pharyngeal flaps from the statistical analyses at the respective ages did not change the results.

VPC-Rate. In contrast to the results for VPC-Sum, VPC-Rate did not differ significantly between IA and NA children at age 5 years ($p = 0.492$) or age 10 years ($p = 0.264$). The results did not change when children who received pharyngeal flaps before the assessments were excluded from the statistical analyses. The proportion of children judged with incompetent VPF decreased in both groups regardless of the method used to evaluate VPF. The results for VPC-Sum and VPC-Rate are shown in Figure 1. Changes in the assessments of VPF between ages 5 and 10 years are shown in Tables 5–8

PCC

The PCC scores at word level at age 5 years did not differ significantly between IA and NA children ($U = 180.00$, $z = -1.86$, $p = 0.063$). After excluding one outlier with significantly lower PCC (NA), a recalculation changed the result to a significant difference ($U = 158.00$, $z = -2.16$, $p =$

TABLE 4 Inter- and intra-transcriber agreement at word and sentence levels at ages 5 and 10 years: median percentages (minimum–maximum)

Age (years)	Agreement on	Inter-transcriber agreement		Intra-transcriber agreement	
		Word level	Sentence level	Word level	Sentence level
5	Correct/incorrect articulation	88% (69–98)	89% (71–100)	95% (84–100)	91% (83–100)
	Symptoms of VPI	85% (49–97)	77% (47–100)	95% (81–100)	87% (74–100)
	Presence of non-oral articulation	96% (76–100)	95% (74–100)	98% (82–100)	99% (83–100)
10	Correct/incorrect articulation	97% (73–100)	97% (74–100)	98% (85–100)	100% (89–100)
	Symptoms of VPI	95% (56–100)	97% (54–100)	92% (74–100)	94% (74–100)
	Presence of non-oral articulation	100% (90–100)	100% (100)	100% (95–100)	100% (97–100)

Note VPI, velopharyngeal insufficiency.

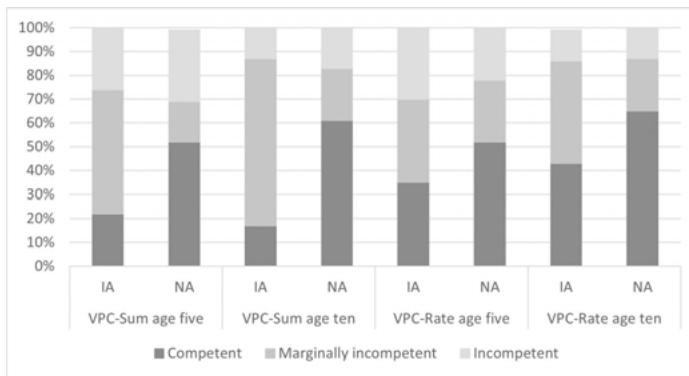


FIGURE 1 Velopharyngeal function at ages 5 and 10 years for internationally adopted (IA) and non-adopted (NA) children.

0.031). At sentence level, the difference was significant ($U = 156.50$, $z = -2.38$, $p = 0.017$).

At age 10 years, there was a significant difference regarding PCC at word level ($U = 164.50$, $z = -2.27$, $p = 0.023$) between IA children and NA children, but not at sentence level ($U = 212.50$, $z = -1.25$, $p = 0.212$). A recalculation after excluding outliers with significantly lower PCC, one in each group for PCC at word level and two in each group for PCC at sentence level did not change the results (Figure 2).

The development of PCC at word and sentence levels between ages 5 and 10 years was significant in both groups (IA: $p < 0.000$ and < 0.000 , respectively. NA: $p = 0.001$ and 0.003 , respectively). If scores within 1 SD (91%) from the age-specific norms are regarded as age-appropriate, 17% of IA children reached this level at age 5 years. The corresponding proportion among NA children was 35%. A total

of 57% of IA children and 39% of NA children scored > 4 SDs below the mean score in typically developed children without CLP. At age 10 years, 30% of IA children and 65% of NA children performed at an age-appropriate level if -1 SD is accepted (95.5%), but 30% in the IA group and 9% in the NA group performed < 4 SDs below the age-specific norm.

Consonant errors

There was no significant difference between IA and NA children regarding CSCs at age 5 years ($p = 0.065$). Oral retracted articulation and non-oral articulation were more common in the IA group (43% and 52%, respectively) than in the NA group (26% and 30%, respectively), but not at

TABLE 5 Changes in the assessment of velopharyngeal function in internationally adopted children between 5 and 10 years of age using sum, score for velopharyngeal competence

	VPC-Sum at age 10			Total
	0	1	2	
VPC-Sum at age 5	0	1	4	5
	1	3	8	12
	2	0	4	6
Total	4	16	3	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Sum, score for velopharyngeal competence; VPF, velopharyngeal function.

TABLE 6 Changes in the assessment of velopharyngeal function in internationally adopted children between 5 and 10 years of age using rating of velopharyngeal competence

	VPC-Rate at age 10			Total
	0	1	2	
VPC-Rate at age 5	0	5	3	8
	1	3	4	8
	2	2	3	7
Total	10	10	3	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Rate, rating of velopharyngeal competence; VPF, velopharyngeal function.

TABLE 7 Changes in the assessment of velopharyngeal function in non-adopted adopted children between 5 and 10 years of age using Sum, score for velopharyngeal competence

	VPC-Sum at age 10			Total
	0	1	2	
VPC-Sum at age 5	0	8	4	12
	1	2	0	4
	2	4	1	7
Total	14	5	4	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Sum, score for velopharyngeal competence; VPF, velopharyngeal function.

a significant level ($p = 0.353$ and 0.231 , respectively). An in-depth analysis of non-oral error types showed that substitution or reinforcement with glottal plosive was more common in IA children (52%) than in NA children (17%),

and this difference was significant ($p = 0.029$). Active nasal fricative was less common in the IA group (4%) than in the NA group, but the difference was not significant (13%, $p = 0.608$).

TABLE 8 Changes in the assessment of velopharyngeal function in non-adopted children between 5 and 10 years of age using rating of velopharyngeal competence

	VPC-Rate at age 10			Total
	0	1	2	
VPC-Rate at age 5	0	8	2	12
	1	4	1	6
	2	3	0	5
Total	15	5	2	23

Notes: 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Rate, rating of velopharyngeal competence; VPF, velopharyngeal function.

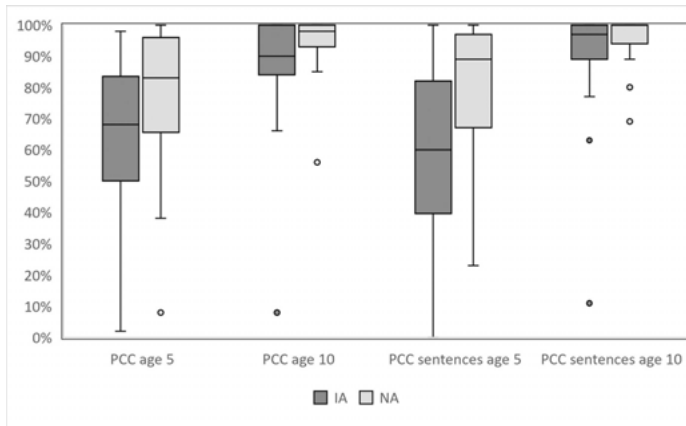


FIGURE 2 Median per cent consonants correct (PCC) at word level and sentence level at ages 5 and 10 years for internationally adopted (IA) and non-adopted (NA) children.

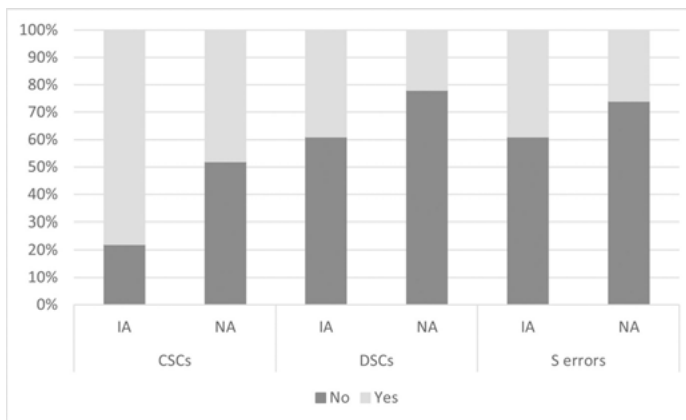


FIGURE 3 Proportions of internationally adopted (IA) and non-adopted (NA) children with articulation errors divided into cleft speech characteristics (CSCs), developmental speech characteristics (DSCs) and s-errors at age 5 years.

At age 5 years, DSCs did not differ significantly between IA and NA children ($p = 0.337$) nor did s-errors ($p = 0.265$). When comparing subtypes of DSCs, velar fronting (IA 22%, NA 13%, $p = 0.699$), stopping (IA 30%, NA 17%, $p = 0.491$), and voicing errors (IA 9%, NA 4%, $p = 1.0$), no significant differences were found between the groups (Figure 3).

At age 10 years, CSCs differed significantly between the groups ($p = 0.023$). Oral retracted articulation remained in 26% of IA children and 9% of NA children ($p = 0.243$), while non-oral articulation was still present in 22% of IA children and 4% of NA children ($p = 0.187$). Glottal plosive was present in 13% of IA children and 0% of NA children ($p = 0.233$), while active nasal fricative was present in 17% of IA children and 4% of NA children ($p = 0.346$).

Regarding DSCs, two IA children still had minor difficulties at age 10 years, one with voicing errors and one with stopping. s-errors did not differ significantly between the groups ($p = 0.414$) (Figure 4).

Secondary palatal surgery

For details on secondary palatal surgeries, see Table 9. One NA child had a re-repair of the soft palate due to dehiscence at age 1;5 and later received a pharyngeal flap (age 3;4). The difference between the proportion of IA and NA children receiving speech-improving surgery was not significant ($p = 0.530$).

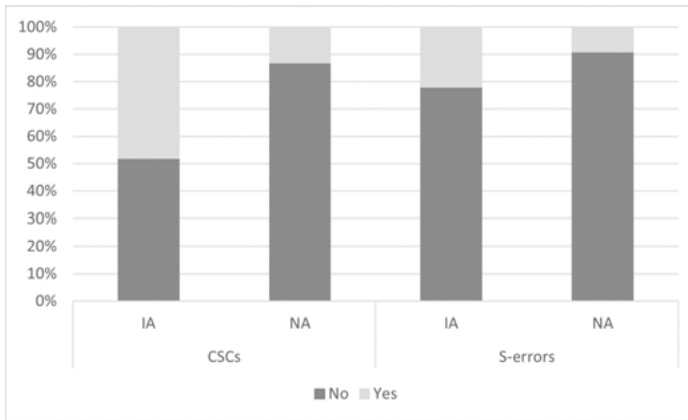


FIGURE 4 Proportions of internationally adopted (IA) and non-adopted (NA) children with articulation errors divided into cleft speech characteristics (CSCs) and s-errors at age 10 years. Only two IA children had developmental speech characteristics (DSCs) (not shown).

TABLE 9 Secondary palatal surgeries

	Internationally adopted	Non-adopted
Secondary surgery due to dehiscence, <i>n</i> (age)	–	1 (1;5)
Fistula repair, <i>n</i> (mean age, SD, range)	4 (3;9, 10, 3;1–4;10)	3 (3;5, 11, 2;5–4;2)
Pharyngeal flap < age 5 years, <i>n</i> (mean age, SD, range)	5 (4;2, 8, 3;3–4;9)	2 (3;8, 6, 3;4–4;1)
Pharyngeal flap age 5–10 years, <i>n</i> (mean age, SD, range)	4 (6;5, 13, 5;3–7;7)	4 (6;2, 17, 5;3–8;3)

Note: Mean ages and ranges are given as years;months, SD is given in months.

Visits to SLP

Mean number of visits to an SLP at age 5 years was 19 (range = 4–35, *n* = 20) in the IA group and 14 in the NA group (range = 3–30, *n* = 18). Between 5 and 10 years of age, the mean number of visits was 12, 5 (range = 2–44, *n* = 21) in the IA group and 8 in the NA group (range = 1–31, *n* = 18). No significant differences were detected between the groups at either age (*p* = 0.450 and 0.335, respectively).

DISCUSSION

This study investigated how articulation and VPF developed longitudinally between ages 5 and 10 years in IA and NA children. Also, it compared VPF, articulation, the proportion of secondary palatal surgery and number of visits to an SLP between these groups. It adds new knowledge of how articulation and VPF develop over time in IA children and how their speech ability is at an older age than previously described. The study demonstrated that developmental articulation errors had ceased at age 10 years in both groups. However, cleft-related articulation errors

were still present in significantly more IA than NA children, and despite the significant development noted, the IA children as a group did not catch up.

Based on previous studies, a somewhat unexpected finding was that the proportion of IA children with incompetent VPF at age 5 years in this study was smaller than reported earlier (Larsson et al., 2020; Scholin et al., 2020). In past studies, as many as half of IA children had incompetent VPF compared with 30% in the present study. The difference between IA and NA children with incompetent VPF was also considerably larger in past studies than in the present study. The proportion of NA children assessed to have incompetent VPF was similar in all three studies, indicating that VPF was assessed with a similar definition of the scale steps, and differences in the assessment can, therefore, not explain the difference between the studies. Further, mean age at arrival to Sweden, at SPC and HPC, and the proportions of secondary surgery were also similar in the studies and cannot explain the difference between them. The three studies differed concerning included cleft types. Larsson et al. (2020) included only children with UCLP, while Scholin et al. (2020) included children with UCLP, BLCP, and isolated cleft palate. How-

ever, it is not certain that this can explain the different outcomes regarding VPF.

Another interesting finding was the discrepancy between VPC-Sum and VPC-Rate, especially in IA children at age 10 years. VPC-Sum revealed a significant difference between the groups at both ages, while VPC-Rate did not even show a trend towards a significant difference. Lohmander et al. (2017a) reported the level of agreement between the two measurements to be 65.2%, in line with what was found in this study. As the VPC-Rate is based on connected speech, which is more like speech in real-life, it is considered to have high external validity. However, similar proportions of children with incompetent VPF in both groups at both ages were identified using both measurements, and the difference lay in the distribution of competent and marginally incompetent VPF. VPC-Sum turned out to be more sensitive than VPC-Rate. This is probably explained by the fact that VPC-Sum is based on transcriptions of target consonants where occurrences of passive and active speech deviations are counted, and VPC-Rate is based on an overall assessment of connected speech. Regardless of the method used to evaluate VPF, those rated with competent VPF were noticeably fewer in the IA group at both ages. With more children in both groups receiving speech-improving surgery, VPF improved between 5 and 10 years.

Children in the IA group scored lower PCC than NA children at both 5 and 10 years, at word and sentence levels. However, most IA and NA children scored below an age-appropriate level. For IA children at ages 5 and 7–8 years, it has previously been reported that only one child scored an age-appropriate PCC, and 16 of the 17 participants scored > 2 SD below the age-specific norm. Further, at ages 7–8 years, most children scored > 4 SD below the age-specific norm (Larsson et al., 2022). As many as 57% in the IA group and 35% in the NA group in the present study scored > 4 SD below the mean score in typically developed children without CLP at age 5 years. Despite significant improvement in both groups, only 30% of the IA children and 65% of the NA children scored at an age-appropriate level, indicating that they still had considerable articulatory difficulties compared with peers without CLP. It is reasonable to believe that the extensive articulation difficulties seen in some children in both groups at age 5 years and mainly in the IA group at age 10 years affected intelligibility and, thus, the opportunity to participate in various contexts.

As PCC does not reveal the character of the articulation error, and different articulation errors affect speech perception differently (Sell & Sweeney, 2020), we chose to divide them into different categories for further analysis, as described previously (Larsson et al., 2020; Morgan et al., 2018; Willadsen et al., 2017). Both CSCs and DSCs were

common in both groups at age 5 years, but almost twice as many IA children as NA children presented with some type of CSC and/or DSC. In this study, the proportion of children in each group with some type of CSC (IA 78%, NA 48%) was similar to that reported by Larsson et al. (2020). However, the results differ with respect to DSCs. Larsson et al. (2020) reported that 92% of their IA group and 65% of their NA group presented with articulation errors related to development. In the present study, the corresponding proportions at age 5 years were 39% in the IA group and 22% in the NA group. A difference in voicing errors largely explained this discrepancy. A total of 76% of IA children and 55% of NA children presented with such errors in the study by Larsson et al. (2020), compared with 9% in the IA group and 4% in the NA group in the present study at age 5 years; at age 10 years, only one child (IA) had voicing errors. Voicing errors were reported by Morgan et al. (2018) to be common in both adoptees and non-adoptees at ages 7–8 years, and in the longitudinal study of IA children, 71% still had voicing errors at ages 7–8 years (Larsson et al., 2022). One explanation for the difference between the present study and earlier work (Larsson et al., 2020, 2022; Morgan et al., 2018) may be difficulties differentiating between a voiced plosive and a voiceless plosive articulated with reduced pressure. Another explanation may be that the SLP focuses more on errors related to VPI, for example, weak pressure consonants, or more phonological aspects, for example, voiced plosives.

Substantial development was seen from age 5 to 10 years in both groups, and errors related to development were seen in only two children (IA) at age 10 years. This suggests that even though DSCs have been reported to be present at ages 7–8 years (Larsson et al., 2022; Morgan et al., 2018), these errors are not as persistent as CSCs. Oral retracted articulation is a common articulation error in children with CLP (Lohmander-Agerskov, 1998), especially when delayed HPC is practiced. In the study by Larsson et al. (2017), oral retracted articulation was more common in NA children than in IA children at age 3 years. The fact that NA children had a residual cleft in the hard palate for a more extended period after SPC was given as an explanation for the results. At age 5 years, the situation was reversed, and oral retracted articulation was noted more frequently in the IA group (Larsson et al., 2020). However, one-third also exhibited velar fronting, suggesting an unstable sound system. The trend of IA children more often having oral retracted articulation than their peers were also seen in the present study, and at age 5 years, approximately one-sixth of the IA children simultaneously had velar fronting. At age 10 years, no child exhibited velar fronting, but more IA than NA children still had oral retracted articulation, suggesting that this articulation error was more persistent in IA children. Unfortunately, about a quarter of the IA



children also still had issues with non-oral articulation, which is associated with VPI and impairs intelligibility even more than retracted oral articulation (Lohmander-Agerskov, 1998). As the IA children had their surgery later than the NA children, the articulation pattern may already have been established at the time of the first surgery, possibly making it more challenging to correct. As shown earlier (Larsson et al., 2017, 2020; Morgan et al., 2018; Scherer et al., 2018; Scholin et al., 2020), the results of this study indicated that IA children have more articulation errors than NA peers with CLP. Further, the results suggested that articulation errors related to the cleft are more persistent in IA children, remaining well into school age. Considering the articulation difficulties seen in both groups, one could have expected to see that reflected in the number of visits to an SLP. However, the mean number of visits to an SLP in both groups was relatively low, especially between ages 5 and 10, and did not differ even though more IA children had articulation difficulties. Providing more intervention may not always be possible since it depends partly on the child's maturity, ability and willingness to participate, and accessibility to an SLP. However, as IA children seem at a higher risk of more persisting and severe articulation difficulties than NA children, it is crucial, when needed, to instigate therapy early and not have a "wait and see" approach. Many children, not least in the IA group, still at the age of 10, had severe speech difficulties. They would probably have benefited from more comprehensive therapy, ideally from an early age, and continued through the early school years.

Our results contradict many of those reported earlier on secondary palatal surgeries, which have been reported to be required considerably more often in IA children than in NA children (Hansson et al., 2012; Morgan et al., 2018; Swanson et al., 2014). Although the proportion was somewhat higher in the IA group in the present study, it did not differ significantly between the groups. This finding should be interpreted cautiously due to the relatively small sample and possible differences in surgical indications between centres. However, differences in surgical results between centres that appear in a subset of challenging-to-operate patients cannot be ruled out.

Several methodological considerations need to be pointed out. In earlier studies, deletion of consonants and replacement with glottal plosive have been reported separately (Larsson et al., 2017, 2020). This is recommended (Sell & Sweeney, 2020), as deletion of consonants can be interpreted as developmental immaturity (Dodd et al., 2003). However, it has also been shown that it is hard to distinguish between consonant deletion and replacement with a glottal plosive (Willadsen et al., 2017). For this reason, we counted each occurrence of consonant

deletion as an elicited target sound and categorized it as an occurrence of glottal plosive.

s-errors occur in just under 30% of Swedish 5-year-olds without cleft palate (Lohmander et al., 2017b), and some earlier studies have reported PCC adjusted for age, with s-errors regarded as correct (Klinto et al., 2014, 2016; Larsson et al., 2020). However, in this study, all s-errors were scored as incorrect. s-errors were present in 39% of IA children and 26% of NA children at age 5 years. The initial aim was to divide s-errors into CSCs and DSCs, but they proved hard to categorize in some cases. Furthermore, when scrutinizing the transcriptions, it was found that the main transcriber had only used the sign for alveolo-palatal fricative (ç) and never the sign for palatal fricative (ç̥). As the alveolo-palatal fricative is a Swedish phoneme and consequently would have been categorized as a DSC, and the palatal fricative is a common CSC, the decision was made not to categorize s-errors as CSCs or DSCs, but to report them separately.

Another methodological consideration in this study is the lack of information on hearing status. Children with cleft palate often exhibit otitis media with effusion, associated with mild conductive hearing loss (Flynn et al., 2009), at least at pre-school age. No child in the study had a severe hearing impairment requiring a hearing aid, but we cannot rule out that some had mild conductive hearing loss. However, the impact of hearing on speech outcomes is sparsely studied. Mild hearing loss was demonstrated to impact the presence of canonical babbling at 10 months, and hearing level at age 18 and 30 months was significantly correlated with PCC at age 36 months (Lohmander et al., 2021), indicating mild conductive hearing loss has an impact on the early development of consonant production. On the other hand, hearing was not associated with speech outcomes at age 3 years (Klinto et al., 2014) or age 5 (Fitzpatrick et al., 2021).

An initial aim of this study was to include an assessment of intelligibility, which would have given important information about the impact of different speech deviations. Recording of spontaneous speech was sparse for most children in both groups at age 5 years, often due to the children's unwillingness or inability to elaborate answers despite encouragement from the SLP who met the children or the SLP's inability to understand the children, which put the conversation to a stop. At age 10 years, the figures had improved, but a considerable proportion of recordings still did not include enough spontaneous speech to enable assessment of intelligibility.

The limited sample size and the heterogeneity in the IA group may have affected the results, and as small studies are vulnerable to outliers, the results should be interpreted cautiously. However, the results in this study point

in the same direction as results already reported and can therefore be considered reliable.

CONCLUSIONS

The IA children in the present study did not have VPI to a greater extent than the NA children, but fewer were judged to have a fully competent VPF. Developmental articulation errors seemed to have ceased at age 10 years, also in most IA children. However, both groups presented with developmental articulation errors at age 5 years, and intervention should thus target both developmental and cleft-related articulation errors. It is evident from the results that IA children have greater and more severe articulation difficulties up to the age of 10 and that they probably require more extended intervention than NA children.

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CONFLICT OF INTEREST STATEMENT


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

DATA AVAILABILITY STATEMENT

The datasets generated during and/or analysed during the current study are available from the corresponding author upon reasonable request.

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ENDNOTES

¹Shriberg and Kwiatkowski (1982) originally developed the measure of PCC in conversational speech. A modified version of PCC in single-word samples has been used to assess articulation skills in children born with cleft palate (Lohmander & Persson, 2008; Scherer et al., 2008; Larsson et al., 2020).

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Paper III



The impact of surgical technique and cleft width on the rate of secondary surgery and velopharyngeal function

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Abstract

Objective: To evaluate different types of intra-velar veloplasty within an otherwise uniform surgical protocol and to examine the impact of cleft width and the surgeon's experience on outcome measurements.

Design: A cross-sectional study.

Setting: A single multidisciplinary craniofacial team at a university hospital.

Participants: 62 individuals with unilateral cleft lip and palate (UCLP) born between 2000-2015 were included. All underwent a 2-stage palatoplasty at 6 and 24 months but were divided into three groups based on the surgical technique used.

Main Outcome Measures: Cleft width was measured on dental casts. Blinded speech and language pathologists assessed velopharyngeal function with the composite score for velopharyngeal competence (VPC-Sum) for single words. They rated velopharyngeal function on a 3-point scale (VPC-R) in sentence repetition. Target sounds in words were phonetically transcribed. The percent of correct consonants (PCC) was calculated.

Results: Cleft width but not surgical technique was significantly associated with the rate of secondary palatal surgery and risk of velopharyngeal insufficiency when using VPC-R. Cleft width was not significantly associated with PCC, and PCC did not differ between the surgical techniques. It appears that improved surgical experience can lead to better outcomes.

Conclusions: Radical muscle dissection did not show superiority over intra-velar veloplasty reinforced by the palatopharyngeal muscle. Longitudinal studies are necessary to evaluate and compare surgical techniques accurately. Cleft width had a more significant impact on the rate of secondary surgery and velopharyngeal function than surgical technique, but neither affected PCC. Further studies should investigate other variables that may affect articulation ability.

Keywords: surgical technique, radical muscle dissection, cleft width, velopharyngeal function, unilateral cleft lip and palate

Introduction

Cleft palate surgery aims to facilitate normal speech and efficient velopharyngeal function, leading to improved articulation. Despite ongoing research, the methods for achieving optimal results still need to be determined. Several factors have been suggested to influence speech outcome and the secondary palatal surgery rate, including the type of cleft (1-3), the cleft's extent in isolated cleft palates (4-6), and cleft width (6-8). In addition to internal factors, various external factors, including the patient's age at the time of the procedure (9-11), the surgeon's experience (12, 13), and the chosen surgical technique (14, 15), are also considered to influence the outcome.

Radical muscle dissection has been suggested to improve velopharyngeal function and articulation while also reducing secondary palatal surgery rates compared to other techniques (16-18). However, comparing the effectiveness of various surgical techniques is difficult due to the above-described multiple factors affecting outcomes as well as differences in age at surgery or assessment.

The present research aimed to assess and contrast different types of intra-velar veloplasty within an otherwise uniform surgical protocol and to investigate the impact of cleft width and the surgeon's experience on outcome measurements. The following research questions were posed: 1) Is radical muscle dissection more effective than intra-velar veloplasty reinforced by the palatopharyngeal muscle in terms of a) reducing the need for secondary palatal surgery due to dehiscence, fistula, or velopharyngeal insufficiency (VPI), b) improving velopharyngeal function (VPF), and c) increasing percent of correct consonants (PCC)? Furthermore, does cleft width or the surgeon's experience affect these results?

Methods

The Regional Ethics Committee in Uppsala approved the study (Reference no.: 2017/457).

Participants

A consecutive series of 109 individuals with unilateral cleft lip and palate (UCLP) born in 2000-2015 and treated at our unit from birth were reviewed at five years of age. Ninety individuals met the inclusion criteria of being non-syndromic and having undergone a 2-stage palatal surgery with soft palate closure at six months (-2, +3 months) and hard palate closure at 24 months (+/-3 months). Twenty-eight of these individuals were excluded due to 1) missing or incomplete data (n=22) or 2) having an additional diagnosis that may affect speech and/or language development (n=6). The study finally included 62 individuals, 19 girls and 43 boys. The initial inclusion criteria were

not met by 19 individuals who had either not followed our surgery protocol, relocated elsewhere, had a syndrome, or were deceased.

Information regarding treatment with tympanostomy tubes was available in 60/62 cases (97%). Forty-two children (68%) received tympanostomy tubes at the time of their lip- or palate surgery. Four children (6%) had tympanostomy tube insertion at their home hospital. Fourteen (23%) were not treated with tympanostomy tubes before five years of age.

The participants were divided into groups based on surgical technique. Due to the slight difference in technique in the group receiving radical muscle dissection, they were split into two groups. Consequently, there were three groups: the non-dissection group, the transition group, and the Sommerlad group.

Surgical treatment

Since 1984, a 2-stage protocol has been used with soft palate closure at six months and hard palate closure at two years. Before 2007, the soft palate was closed with an intra-velar veloplasty reinforced by the palatopharyngeal muscle (19) (non-dissection group). In this technique, intra-velar muscle repositioning was achieved by releasing the muscle and nasal layer from the posterior edge of the hard palate without separating the levator muscle from the nasal mucosa. Radical muscle dissection was then introduced in 2007 and made progressively more radical (transition group). Since August 2009, radical muscle dissection, as described by Sommerlad (12, 20), has been performed consistently (Sommerlad group). In all cases, incision lines were placed at the borders of the cleft without releasing incisions, but usually with an anterior-based vomerian flap to the nasal layer of the soft palate. Four surgeons have been involved during the study period; see Table 1. Surgeon 1 and 2 performed the soft palate closure without loop magnification, while Surgeon 3 and 4 performed it under 3.5 loop magnification. Since 2014, all soft palate closures have been carried out by Surgeon 3 and 4 under a microscope with 2.3-14 times magnification. Usually, 6-10 times magnification is used.

Table 1. The number of soft palate surgeries grouped by surgeon and technique.

Technique	Surgeon 1	Surgeon 2	Surgeon 3	Surgeon 4
Non-dissection (n)	6	3	7	
Transition (n)		4	10	
Sommerlad (n)			19	13

Results regarding secondary palatal surgery and VPF rates, categorized by the type of surgery, will only be reported for the surgeon who has been active throughout the study period.

The residual cleft in the hard palate was repaired in two layers at two years of age. If needed for closure without tension, releasing incisions were

performed, as described by von Langenbeck (21). When using releasing incisions, the incision lines were always placed along the cervical lines of the teeth.

Speech-improving surgery was performed with a re-repair of the soft palate (only in children initially operated according to Sommerlad) or a pharyngeal flap. Before such surgery, the patients were evaluated with a speech assessment, nasoendoscopy, and, when possible, nasometry.

Cleft width measurement

A dental study cast was obtained at the time of the lip-plasty at three months. The casts were analyzed to determine cleft width using reference points and linear measurements earlier described (22-24). Using a digital caliper, the cleft width was calculated as the ratio A-A1/T-T1 and measured to the nearest 0.01 mm. T-T1 represents the posterior width of the alveolar arch in the tuber area, and A-A1 represents the width of the cleft at the level of T-T1. Thirty percent of the casts were randomly chosen for repeated measurements to establish intra-rater agreement. Dental casts were available for 56 individuals; also included were measurements for six additional individuals whose dental casts had been previously measured in a previous study (8) using the same methodology and measurer as in the present study.

Speech material

A speech sample was recorded at the routine follow-up at five years of age. The audio recordings were made with Zoom H4n or a PC with Soundswell software (Saven Hitech, Stockholm, Sweden) and a condenser microphone (Røde NT4, Sydney, Australia or Philips SpeechMike Classic 6264Details) using the Swedish Articulatory and Nasality Test (SVANTE) (25). Three sets of speech material were edited from the recordings using Praat software (26): 1) Fifty-nine single words, each including one target consonant; 2) A 9-word string (the first nine monosyllabic words edited to a string of words with no pause between them) including high vowels for assessment of hypernasality; 3) Twelve sentences for perceptual assessment (ten including oral consonants, two including oral and nasal consonants) of velopharyngeal competence (VPC-R). All edited audio files were randomized and assigned codes to enable blinded assessments.

Speech assessment

Experienced speech and language pathologists (SLPs), two independent SLPs (Rater 1 and 2), and the first author (Rater 3) rated hypernasality and VPC-R. One of the independent SLPs (Rater 2) and the first author (Rater 3) also carried out phonetic transcriptions of target consonants in words. Before the

speech assessments, a calibration session was conducted to ensure as equivalent assessments as possible.

Two different measurements for assessing VPF were used: the rating of perceived velopharyngeal competence in sentences (VPC-R) and a composite score for velopharyngeal competence (VPC-Sum) for single words (27, 28). The SLPs rated VPF on a 3-point ordinal scale: 0 = competent, 1 = marginally incompetent, and 2 = incompetent. The score for each child was classified based on the majority decision of the three SLPs' ratings. In case of total disagreement, the middle value was chosen.

VPC-Sum is a composite score including a) perceptual ratings of hypernasality rated on a 4-point ordinal scale (0 = normal, 1 = mild, 2 = moderate, and 3 = severe), b) perceptual signs of VPI from transcriptions (audible nasal air leakage, weak pressure consonants, and nasal realization of voiced consonants), and c) active non-oral speech errors from transcriptions, used to assess VPF at the word level. Each of the three variables generated a score of 0–2 and was added to calculate VPC-Sum 0-6. In the statistical analyses, the results from VPC-Sum will be used according to the interpretation of VPC-Sum: 0-1 = 0: competent, 2–3 = 1: marginally incompetent, and 4–6 = 2: incompetent.

A semi-narrow phonetic transcription was performed, in the present study defined as a phonetic transcription using the International Phonetic Alphabet (IPA) (29), and the symbols for nasal escape, velopharyngeal friction, weak articulation, voicing, devoicing, and active nasal fricative from the extended IPA symbols for disordered speech (30). The transcriptions carried out by the independent SLP were used as the results for all participants. Percentage of correct consonants (PCC) was developed to evaluate PCC in conversational speech (31), and a modified version of PCC in single words has been used to assess articulation in children with cleft lip and palate (CLP) (32-34). PCC was calculated based on the transcriptions, where all consonant errors were weighted equally. The target consonant was scored as correct if the place and manner of articulation were correct. Signs of VPI were disregarded. The audio files were listened to through high-quality headphones and could be replayed as many times as needed. Thirty percent of the audio files were randomly selected for re-assessment to calculate intra-rater agreement.

Statistical analysis

Single measures intra-class correlation coefficient with a 2-way mixed-effects model (ICC) was used to determine intra-rater reliability for cleft width measurement. The levels of observed agreement were interpreted as follows: <.40 is poor, .40-.59 is fair, .60-.74 is good, and .75-1.00 is excellent (35). Logistic regression analysis was used to calculate the impact of surgical technique and cleft width on the rate of secondary surgery. Ordinal regression analysis estimated the association between surgical technique and cleft width with VPC-

R and VPC-Sum. Spearman correlation analysis was used to detect any association between cleft width and PCC. Kruskal-Wallis was used to detect any differences in PCC between surgical techniques. The results for the surgeon active throughout the study period will be presented descriptively. Statistical analyses were performed in IBM SPSS Statistics, version 29, or R. The level of significance was set at $\alpha < 0.05$ (2-tailed).

Reliability

Intra-rater agreement for cleft width measurement was good (.686, 95% CI .315-874 and .718, 95% CI .475-.860) respectively). Inter-rater agreement for hypernasality and VPC-R was calculated as the frequency of 1) agreement between all three raters, 2) agreement between two raters, and 3) no agreement. The intra-rater agreement was measured as agreement point by point for both hypernasality and VPC-R, as were the inter- and intra-transcriber agreement for the phonetic transcriptions. Comparisons were made for correctly articulated target sounds, manner of articulation, place of articulation, VPI symptoms, and non-oral articulation. Inter- and intra-rater agreement for hypernasality, VPC-R, and the transcriptions are shown in Table 2 and 3. Minor differences between the transcribers that were considered as an agreement were /ʔ and bʔ/ (glottal vs. glottal reinforcement), /ʔ and Ø*/ (*omission of a consonant), /c and k/, and /ʝ and g/.

Table 2. Inter- and intra-rater agreement for hypernasality and VPC-R.

		Hypernasality	VPC-R
Inter-rater agreement	All three raters agree (%)	48%	57%
	At least two out of three raters agree (%)	92%	98%
	No agreement (%)	8%	2%
Intra-rater agreement	Rater 1	80%	74%
	Rater 2	85%	95%
	Rater 3	70%	100%

VPC = velopharyngeal competence.

Table 3. Inter- and intra-transcriber agreements for transcriptions. Median percentages (min–max).

Agreement on	Inter-transcriber agreement	Intra-transcriber agreement	
		Rater 2	Rater 3
Correct/incorrect articulation	91% (65-100)	95% (85-100)	97% (84-100)
Manner of articulation	98% (76-100)	98% (88-100)	100% (96-100)
Place of articulation	90% (74-100)	95% (77-100)	96% (83-100)
Symptoms of VPI	93% (70-100)	97% (87-100)	97% (86-100)
Presence of non-oral articulation	100% (82-100)	100% (87-100)	100% (89-100)

VPI = velopharyngeal insufficiency.

Results

Surgery

The age at primary surgery, mean cleft width, and the rate of secondary palatal surgery divided into the three groups according to surgical technique are shown in Table 4. Logistic regression analysis showed that surgical technique was not significantly associated with the need for secondary palatal surgery due to dehiscence, fistula or VPI but cleft width was; see Table 5.

Table 4. Primary and secondary surgery.

	Surgical technique		
	Non-dissection, n=16	Transition, n=14	Sommerlad, n=32
Soft palate closure (mean age)	6.8	6.9	6.8
Hard palate closure (mean age)	24.5	24.2	24.4
Cleft width (mean ratio, range)	30% (14-43%)	33% (22-44%)	35% (24-45%)
Dehiscence (soft palate)	1 (6%)	1 (7%)	-
Dehiscence (hard palate)	1 (6%)	1 (7%)	1 (3%)
Fistulas (hard/soft palate junction)	-	-	2 (6%)
Fistulas (alveolar)	1 (6%)	-	-
Speech-improving surgery	1 (6%)	2 (14%)	3 (9%)*
Secondary palatal surgery (total)	4 (25%)	3 (21%)	5 (16%)

Age at surgery is given in months. *One with pharyngeal flap, two with re-repair of the soft palate.

Table 5. Logistic regression analysis on how surgical technique and cleft width were associated with the need for secondary surgery.

Variable	OR	95% CI	p
Non-dissection (n=16)	REF		.361
Transition (n=14)	.659	.106-4.093	.655
Sommerlad (n=32)	.309	.059-1.621	.165
Cleft width	1.141	1.021-1.275	.020

Speech

The SLPs' assessment showed that a total of six individuals had VPI when using VPC-R and eight when using VPC-Sum. To accurately represent the results of the surgical techniques used, we looked at the primary VPI (6, 36). The primary VPI included individuals with a VPC-R score of 2 or a VPC-Sum score of 4 or higher from the SLPs' assessment combined with those who underwent speech-improving surgery before age five. The SLPs' assessments and the primary VPI are shown in Table 6.

In the statistical analyses, all individuals who had speech-improving surgery were counted as having VPI (VPC-R = 2 and VPC-Sum = 4-6) regardless of the assessments. Ordinal regression analysis showed no significant association between VPC-R and the surgical technique used, but VPC-R was significantly associated with cleft width. Neither surgical nor cleft width was associated with VPC-Sum; see Table 7.

There was no significant correlation between cleft width and PCC ($\rho = -.128, p = .329$). A Kruskal-Wallis calculation found no differences in PCC between surgical techniques ($H(2) = .070, p = .966$).

Table 6. Those assessed as having velopharyngeal insufficiency (VPI) at the 5-year assessment and primary VPI (those assessed with VPI plus those who had speech-improving surgery before the 5-year assessment).

	VPC-R		VPC-Sum	
	VPC-R = 2 N (%)	Primary VPI N (%)	VPC-Sum \geq 4 N (%)	Primary VPI N (%)
Non-dissection (n=16)	1 (6%)	2 (13%)	2 (13%)	3 (19%)
Transition (n=14)	3 (21%)	4 (29%)	5 (36%)	5 (36%)
Sommerlad (n=32)	2 (6%)	5 (16%)	1 (3%)	4 (13%)

VPC = velopharyngeal competence.

Table 7. Ordinal regression analyses on how surgical technique and cleft width were associated with VPC-R and VPC-Sum.

Variable	VPC-R			VPC-Sum		
	OR	95% CI	<i>p</i>	OR	95% CI	<i>p</i>
Non-dissection (n=16)	REF			REF		
Transition (n=14)	3.448	.663-17.932	.141	3.784	.731-19.585	.113
Sommerlad (n=32)	.814	.164-4.039	.801	1.227	.270-5.566	.791
Cleft width	2.700	1.053-6.919	.039	1.985	.845-4.662	.116

Impact of surgeon's experience

Results divided by technique for the surgeon active throughout the study period are displayed in Figure 1 a-c.

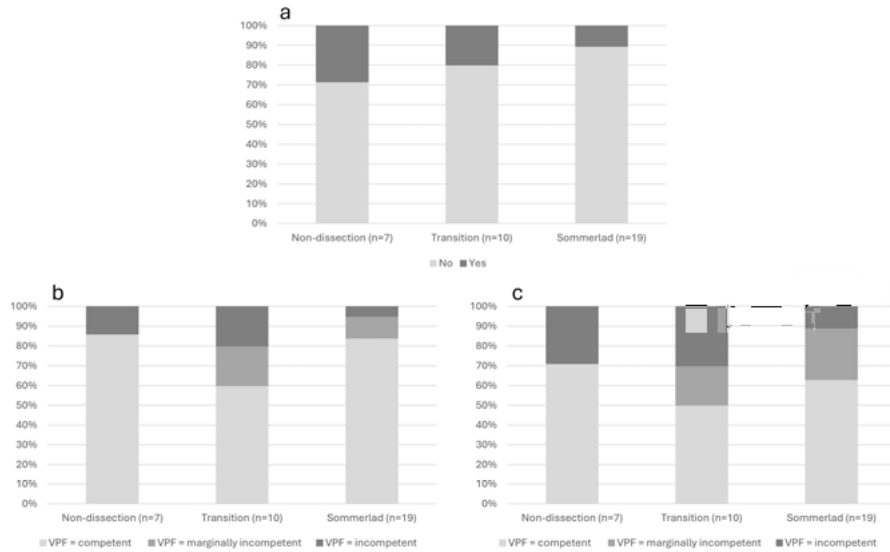


Figure 1. a) The rate of secondary palatal surgery, b) Velopharyngeal function (VPF) assessed with VPC-R, and c) VPF assessed with VPC-Sum divided into surgical techniques for the surgeon active throughout the study period. Those who had undergone speech-improving surgery before the assessment are counted as having velopharyngeal insufficiency.

Discussion

The present study analyzed the impact of radical intra-velar veloplasty within an otherwise uniform surgical protocol for palatal reconstruction and investigated the effect of cleft width and the surgeon's experience. In 2007, the radical intra-velar veloplasty technique was introduced and performed progressively more radically. Then, in late 2009, radical intra-velar veloplasty, as described by Brian Sommerlad (12, 20), was adopted. Therefore, the group undergoing radical muscle dissection was divided into two groups to demonstrate any differences between the two well-described techniques as well as to identify any effects of the transition period. The study found that the rate of secondary palatal surgery and risk of VPI when using VPC-R were determined by cleft width rather than surgical technique. Neither was associated with the risk of VPI when using VPC-Sum. No correlation was found between cleft width and PCC, and no significant difference was observed between techniques for PCC.

Logistic regression showed no significant link between surgical technique and the rate of secondary palatal surgery. Repair of fistulas occurring at the junction between the soft and hard palate was observed only in the Sommerlad group, with two individuals (6%) having undergone surgery. This finding is consistent with other studies reporting similar rates of fistula occurrence requiring surgical intervention in individuals with UCLP after radical intra-velar veloplasty following Sommerlad's technique (6, 37).

The surgical technique did not affect VPF outcomes, but patients who underwent surgery during the transition period had higher VPI rates than the other two groups. In the Scandcleft studies, they cautioned that new and complex techniques might potentially harm (13, 28). This is evident during the transition period but not after the technique described by Sommerlad (12, 20) was adopted, where the initial results were comparable to the later ones. As radical intra-velar veloplasty had been used for over two years when the Sommerlad technique was adopted, the surgeon's increased experience could account for this. Still, it may also be because the surgeon who introduced the Sommerlad technique at our unit underwent training from Brian Sommerlad prior to the implementation of this technique. This highlights the importance of adhering to a systematic process with structured training when introducing new surgical techniques.

In the present study, the primary VPI rates for the Sommerlad technique were 16% (VPC-R) and 13% (VPC-Sum). Doucet et al. (17) reported VPI in 15% at a mean age of 3:3 years; Dissaux et al. (18) reported VPI in 14% at five years of age, and Klintö et al. (37) reported VPI in 15% at five years of age. Our results are similar to those mentioned above but higher than the 5.2% reported by Baillie and Sell (6). However, our group had fewer participants, which may have had a greater impact on the results. Another possible explanation may be individual differences within and between surgeons in executing the technique. Indeed, during interviews with fourteen surgeons regarding execution of the Sommerlad technique, it was discovered that there was sometimes variation in execution due to, for example, patient heterogeneity or differences in training experience (38).

Research has shown that cleft width is associated with the need for more secondary palatal surgery and VPI (6-8), and the present study partly supports that finding. The cleft width was significantly associated with the requirement for secondary palatal surgery, and individuals with wider clefts were more likely to have VPI when using VPC-R. The mean cleft width in the non-dissection group was narrower than in the Sommerlad group. Somewhat contradictory in this regard is the greater rate of secondary surgery in the non-dissection group, suggesting that surgical technique may also play a role, although evidence for this was not found in the present study.

One limitation of the present study is the involvement of four surgeons, which may lead to individual differences in evaluating the techniques due to their different surgical experiences and skills. However, upon examining the

results of the only surgeon who was active throughout the entire study period, it became evident that the use of the Sommerlad technique resulted in better outcomes in relation to both the rate of secondary palatal surgery and VPI. Although one might assume that the introduction of the Sommerlad technique would have led to worse results initially, it is crucial to note that radical muscle dissection, although not as radical as in the Sommerlad technique, had already been utilized for over two years. Therefore, any improvements observed after the transition period can possibly be attributed to increased experience.

Another limitation of the study is the sample size. The study's sample size could have been increased if the age range for the operations had been widened. However, this would have raised the question of whether age at surgery impacted the outcome.

Six dental casts were missing, and the cleft width measurements for those individuals were taken from a previous study. Because the same method and measurer were utilized in the present study, this was deemed to have no impact on the results.

Conclusion and clinical implications

When a new surgical technique is introduced, the aim is to improve speech and reduce the burden of care. While the Sommerlad technique did not show significant superiority over the non-dissection technique regarding speech outcomes, the rate of secondary palatal surgery decreased from 25% to 16%. However, long-term studies are needed to evaluate and compare surgical methods accurately, as final treatment results can only be assessed once patients have finished growing.

A systematic process with structured training is paramount when introducing new surgical techniques.

In the present study, cleft width was found to have a more significant impact than radical intra-velar veloplasty, with wider clefts being more vulnerable to secondary surgery and VPI. However, neither cleft width nor surgical technique affected PCC, and future studies should consider other variables that may affect articulation ability.

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Paper IV



Associations between cleft type and width and the rate of secondary palatal surgery and articulation proficiency in 5-year-olds with cleft lip and/or palate.

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Abstract

Objective: To investigate the association of cleft type and width with the rate of secondary palatal surgery, articulation, and velopharyngeal function (VPF).

Design: A cross-sectional study.

Setting: A single multidisciplinary craniofacial team at a university hospital.

Patients: 100 patients with a non-syndromic cleft lip and/or soft and hard palate born between 2000 and 2015 and treated with a 2-stage palatoplasty were included. Twenty-one had cleft in the soft and hard palate (SHCP), 17 had bilateral cleft lip and palate (BCLP), and 62 had unilateral cleft lip and palate (UCLP).

Main outcome measures: The impact of cleft type and width on the rate of secondary palatal surgery, the percent of correct consonants (PCC), and PCC adjusted for age (PCC-A), and the composite score for velopharyngeal competence (VPC-Sum) at five years of age. Articulation errors were divided into cleft speech characteristics (CSCs) and developmental speech characteristics (DSCs), and the types of errors were compared between the groups.

Results: Neither cleft type nor cleft width was associated with the rate of secondary surgery or VPC-Sum. Cleft width but not cleft type was significantly associated with PCC and PCC-A. Both CSCs and DSCs were more common in the BCLP group but not at a significant level. The types of errors did not differ between cleft types.

Conclusions: Cleft width predicted PCC and PCC-A and should be included in analyses to identify factors that may impact different outcomes. Cleft type does not seem to be a reliable predictor.

Keywords: Cleft width, cleft type, secondary surgery, articulation, PCC, PCC-A

Introduction

Cleft type is considered one of several factors influencing the rate of secondary palatal surgery (1) and speech outcomes (2). Other factors, such as cleft width (3, 4), timing of repair (5-8), and extent of cleft in isolated cleft palates (2, 9-11), are also thought to influence surgical and speech outcomes. It is common to consider all cleft types when examining surgical outcomes. However, regarding speech outcomes, studies often focus on a single selected cleft type. In recent years, there has been an increase in studies that include all types of cleft palate when examining speech outcomes.

A meta-analysis showed strong evidence that clefts involving only the soft palate were associated with less secondary palatal surgery than other cleft types and that bilateral cleft lip and palate (BCLP) were associated with more secondary palatal surgery (1). It has been found through various studies that wider clefts are more likely to require secondary palatal surgery (12-14). Studies controlling for both cleft type and cleft width have demonstrated that cleft width is associated with the rate of such surgery rather than cleft type itself (4, 15).

Individuals with BCLP have been shown to have significantly poorer articulation than those with cleft palate only (CPO) and unilateral cleft lip and palate (UCLP); that is, the more extensive cleft, the more articulation difficulties (2, 10, 16-20), but not necessarily significantly more signs of velopharyngeal insufficiency (VPI) (16, 17, 19). It has been shown that individuals with a cleft in both the soft and hard palate have poorer speech outcomes than those with clefts in only the soft palate (2, 9-11). The studies demonstrating that individuals with BCLP have poorer articulation than other cleft types did not control for cleft width when assessing articulation ability.

This study aimed to investigate the association between cleft type and width and the rate of secondary surgery and speech outcomes in terms of articulation and velopharyngeal function (VPF). The following research questions were posed: 1) Is cleft type and width associated with the secondary surgery rate due to dehiscence, fistula, or VPI? 2) Is cleft type and width associated with articulation and VPF? 3) Are there differences regarding the type of articulation errors between cleft types?

Methods

The Regional Ethics Committee in Uppsala approved this study (Reference no.: 2017/457).

Participants

A consecutive series of 356 children born 2000-2015 with cleft lip and or soft and hard palate and treated at our unit from birth were reviewed at five years

of age. One-hundred and fifty-five met the inclusion criteria of being non-syndromic, treated at our unit from birth, and treated according to a surgical protocol, including a 2-stage palatal surgery with soft palate closure at six months (-2, +3 months) and hard palate closure at 24 months (+/-3 months). Fifty-five children were excluded due to 1) missing or incomplete data (n=40) or 2) an additional diagnosis that may affect speech and language development (n=15). A total of 100 children were finally included: 62 with UCLP (19 girls and 43 boys), 17 with BCLP (six girls and 11 boys), and 21 with a cleft in the soft and hard palate (SHCP) (15 girls and six boys).

Information about treatment with tympanostomy tubes was available for 89% of the cases. A total of 65 children (65%) received tympanostomy tubes simultaneously with lip- or palate surgery, (42/62 (68%) with UCLP, 12/17 (71%) with BCLP, and 11/21 (52%) with SHCP). An additional seven children had notes of tympanostomy tube insertion at their home hospital (four with UCLP, two with BCLP, and one with SHCP). Seventeen had not been treated with tympanostomy tubes (fourteen with UCLP, one with BCLP, and two with SHCP).

Information regarding the number of visits to a speech and language pathologist (SLP) was available for 83% (52/62 (84%) with UCLP, 14/17 (82%) with BCLP, and 17/21 (81%) with SHCP), out of which 40 (48%) had received speech intervention (25/52 (48%) with UCLP, 6/14 (43%) with BCLP, and 9/17 (53%) with SHCP). On average, the children had 12 visits ranging from one to 44. Those who had not received speech intervention had an average of 6 visits, ranging from 1 to 16. Those who had received speech intervention had an average of 18 visits ranging from three to 44.

Outcome data on the percent of correct consonants (PCC) and VPF in individuals with UCLP was also included in a study investigating the impact of surgical techniques and cleft width on the need for secondary surgery and velopharyngeal function (21).

The initial inclusion criteria were not met by 201 individuals, of which 111 did not meet the criteria of 2-stage surgery or the age at surgery. Eighty-six of these individuals had SHCP, of which 74 were treated with 1-stage palatoplasty. Additional reasons for being excluded were relocating outside our region or being deceased.

Surgical treatment

The standard procedure at our unit is to perform lip surgery at three months, soft palate closure at six months, and hard palate closure at two years. Until 2007, intra-velar veloplasty reinforced by the palatopharyngeal muscle (22) was performed. Radical muscle dissection was introduced in 2007; since August 2009, it has been performed according to Sommerlad (23, 24). Speech-improving surgery was performed with either a re-repair of the soft palate or a pharyngeal flap. Before speech-improving surgery, all individuals

underwent a speech assessment and a nasoendoscopy. Surgical details for the included individuals are shown in Table 1.

Table 1. Primary and secondary surgery until five years of age.

	Cleft type		
	UCLP (n=62)	BCLP (n=17)	SHCP (n=21)
Soft palate closure (mean age)	6.8	7.3	6.8
Hard palate closure (mean age)	24.3	24.1	24.3
Cleft width (mean ratio, range)	33% (14-45%)	37% (23-45%)	25% (8-39%)
Dehiscence (soft palate)	2 (3%)	1 (6%)	-
Dehiscence (hard palate)	3 (5%)	2 (12%)	3 (14%)
Fistulas (hard/soft palate junction)	2 (3%)	1 (6%)	1 (5%)
Fistulas (alveolar)	1 (2%)	1 (6%)	-
Speech-improving surgery	6 (10%)	3 (18%)	4 (19%)
Secondary palatal surgery (total)	12 (19%)	6 (35%)	6 (29%)

Age at surgery is given in months. The rates of secondary palatal surgeries are provided in the number of individuals and percent. Mean ratio = ratio A-A1/T-T1. UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = cleft in the soft and hard palate.

Cleft width measurement

Dental study casts were obtained at the time of lip-plasty at three months of age for those with UCLP or BCLP and at the time of soft palate closure at six months for those with SHCP. Using reference points and linear measurements earlier described (25-27), an orthodontist with more than fifteen years of experience measured the dental study casts. Using a digital caliper, the cleft width was calculated as the ratio A-A1/T-T1 and measured to the nearest 0.01 mm. T-T1 represents the posterior width of the alveolar arch in the tuber area, and the A-A1 represents the width of the cleft at the level of T-T1. Thirty percent of the casts were randomly chosen for repeated measurements to establish intra-rater agreement. Dental casts were available for 90 individuals. Measurements for the ten individuals whose dental casts were missing were obtained from an earlier study (4) using the same methodology and measurer as in the present study.

Speech material

At the routine control at five years of age, a speech sample was audio-recorded with Zoom H4n or a PC with Soundswell software (Saven Hitech, Stockholm, Sweden) and a condenser microphone (Røde NT4, Sydney, Australia or Philips SpeechMike Classic 6264). The speech material used was the Swedish Articulatory and Nasality Test (SVANTE) (28). Using Praat (29), two sets of speech material were edited from the recordings: 1) Fifty-nine single words, each including one high-pressure target consonant (plosives /p/, /b/, /t/, /d/, /k/, and voiceless fricatives /f/, and /s/ in initial, medial and final position, and

the voiceless fricative /ɸ/ in initial position); 2) A nine-word string (the first nine monosyllabic words edited to a string of words with no pause between them) including high vowels (/i:/, /u:/ and /ɜ:/) for assessment of hypernasality. All edited audio files were randomized and assigned codes to enable blinded assessments.

Speech assessment

The assessments involved three SLPs with at least five years of experience in cleft palate speech, two independent SLPs (raters 1 and 2) plus the first author (rater 3). A calibration session was conducted before the speech assessments to ensure equivalent assessments. Prior to the calibration sessions, all raters and transcribers did their individual ratings of hypernasality and velopharyngeal function (13 audio recordings) and transcriptions (4 audio recordings). During the following calibration sessions, if there were disagreements among the SLPs regarding the ratings or transcriptions, the audio recordings were listened to, and consensus between raters was obtained after discussion. All three SLPs rated hypernasality, while one of the independent SLPs (rater 2) and the first author (rater 3) performed the phonetic transcriptions of target consonants in words.

A semi-narrow phonetic transcription using the International Phonetic Alphabet (30) and the symbols for nasal escape, velopharyngeal friction, weak articulation, voicing, devoicing, and active nasal fricative from the extended IPA (31) symbols for disordered speech was performed by raters 2 and 3. The transcriptions made by the independent SLP were used as the results for all participants. Percent of correct consonants (PCC) was initially developed by Shriberg and Kwiatkowski in 1982 to evaluate PCC in conversational speech (32), and modified versions of PCC in single words have been used to assess articulation in children with CLP (33-35). All consonant errors were weighted equally, and based on the transcriptions, PCC was calculated. The target consonant was scored correct if the place and manner of articulation were correct. In addition, the percent of correct consonants adjusted for age (PCC-A) (36) was calculated. In PCC-A, age-appropriate s-distortions such as inter-dental, lateral, supra-dental, retroflex, alveolo-palatal, and palatal production of /s/ is scored as correct. When calculating PCC and PCC-A, any signs of VPI were disregarded. As described in the Scandleft project (37, 38), articulation errors were divided into cleft speech characteristics (CSCs) and developmental speech characteristics (DSCs). CSCs were subdivided into non-oral errors (glottal or pharyngeal plosive or fricative, nasal for unvoiced stop or fricative, and active nasal fricative) and oral errors (retracted/backed to palatal/velar/uvular place of articulation and double articulation). Errors related to development in the present study were velar fronting (a velar consonant is replaced with a dental or alveolo-dental consonant), stopping (a fricative is replaced with a plosive), and voicing errors (difficulties differentiating between

voiced and voiceless consonants) and were considered DSCs. Three or more occurrences signified an error. An articulation error could be counted as both a CSC and a DSC.

To assess velopharyngeal competence (VPC), VPC-Sum was used (39, 40). VPC-Sum is a composite score including a) perceptual ratings of hypernasality rated on a 4-point scale (0 = normal, 1 = mild, 2 = moderate, and 3 = severe), b) perceptual signs of VPI from transcriptions (nasal emission and weak pressure consonants), and c) active non-oral speech errors from transcriptions, used to assess VPC at the word level. The three variables generated a score of 0–2 and were added to calculate VPC-Sum, as shown in Table 2.

The audio files were listened to through high-quality headphones and could be replayed as many times as needed. Twenty-five percent of the audio files were randomly selected for re-assessment to calculate intra-rater agreement.

Table 2. Transfer rules for values from the assessment of hypernasality, active non-oral errors, and symptoms of velopharyngeal insufficiency (VPI) (audible nasal air leakage, weak pressure consonants, and nasal realization of voiced consonants) into summary scores of the VPC-Sum.

Variables	Transfer rules			Summary
Hypernasality (ordinal 4-point scale)	0 = 0	1 = 1	2–3 = 2	0–2
Active non-oral errors (number from phonetic transcription)	0–2 = 0	3–5 = 1	≥6 = 2	0–2
VPI-symptoms (number from phonetic transcription)	0–2 = 0	3–5 = 1	≥6 = 2	0–2
VPC-Sum*				0–6

*Interpretation VPC-Sum. 0–1 = 0: Competent. 2–3 = 1: Marginally incompetent: evidence of minor problems suggesting borderline closure. 4–6 = 2: Incompetent: evidence of significant problems usually requiring surgical management.

Statistical analysis

To determine intra-rater reliability for cleft width measurement, the single measures intra-class correlation coefficient with a 2-way mixed-effects model (ICC) was used. The levels of observed agreement were interpreted according to Cicchetti (41) as follows: <.40 is poor, .40-.59 is fair, .60-.74 is good, and .75-1.00 is excellent. Logistic regression analysis was used to investigate the impact of cleft type and cleft width on the rate of secondary palatal surgery. The effect of cleft type and cleft width on PCC and PCC-A was analyzed using linear regression analysis. An ordinal regression analysis investigated the association between cleft type and cleft width with VPF. Possible differences between the cleft types regarding CSCs and DSCs were calculated with the Chi-2 test. Statistical analyses were performed in IBM SPSS Statistics, version 29, or R. The level of significance was set at $\alpha < 0.05$ (2-tailed).

Reliability

Intra-rater reliability for cleft width measurement was good (.718, 95% CI .475-.860). Inter-rater agreement for hypernasality was calculated as the frequency of 1) agreement between all three raters, 2) agreement between two raters, and 3) no agreement. The inter- and intra-rater agreement for hypernasality was measured as agreement point-by-point, as was inter- and intra-rater agreement for the transcriptions. Comparisons were made for correctly articulated target sounds, manner of articulation, place of articulation, VPI symptoms, and non-oral articulation. Minor differences were considered an agreement in calculating inter- and intra-transcriber agreement (see Table 3). Inter- and intra-rater agreement for hypernasality and the transcriptions are shown in Table 4 and 5.

Table 3. Differences between phonetic transcriptions considered as agreement.

/ʔ b̥ʔ/ (glottal vs. glottal reinforcement)
/ʔ ø*/
/c k/ /ɟ g/
*omission of target consonant.

Table 4. Inter- and intra-rater agreement for hypernasality.

		Hypernasality
Inter-rater agreement	All raters agree (%)	45%
	At least two out of three raters agree (%)	92%
	No agreement (%)	8%
Intra-rater agreement	Rater 1	77%
	Rater 2	85%
	Rater 3	69%

Table 5. Inter- and intra-transcriber agreements for transcriptions. Median percentages (min–max).

Agreement on	Inter-transcriber agreement	Intra-transcriber agreement	
		Rater 2	Rater 3
Correct/incorrect articulation	91% (58-100)	96% (81-100)	97% (84-100)
Manner of articulation	98% (76-100)	98% (88-100)	100% (96-100)
Place of articulation	89% (65-100)	95% (77-100)	97% (83-100)
Symptoms of VPI	93% (70-100)	97% (79-100)	96% (83-100)
Presence of non-oral articulation	100% (70-100)	100% (85-100)	100% (89-100)

VPI = velopharyngeal incompetence

Results

Secondary palatal surgery

Logistic regression analysis showed that cleft type was not associated with the rate of secondary palatal surgery due to dehiscence or fistula or the rate of speech-improving surgery, nor was cleft width; see Table 6.

Table 6. Logistic regression analyses on how cleft type and cleft width were associated with the rate of secondary palatal surgery.

Variable	Secondary palatal surgery due to fistula or dehiscence			Speech-improving surgery		
	OR	95% CI	<i>p</i>	OR	95% CI	<i>p</i>
UCLP (n=62)	REF		.490	REF		.164
BCLP (n=17)	1.507	.336-6.760	.593	1.515	.322-7.131	.599
SHCP (n=21)	2.435	.535-11.079	.250	4.858	.950-24.850	.058
Cleft width	1.033	.952-1.122	.437	1.099	.998-1.209	.054

UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, OR = odds ratio, CI = confidence interval

Articulation

Linear regression analysis showed that cleft type was not significantly associated with PCC or PCC-A, but cleft width was; see Table 7. Median PCC and PCC-A are shown in Figure 1.

Table 7. Linear regression analysis on how cleft type and cleft width were associated with the percent of consonants correct (PCC) and PCC adjusted for age (PCC-A).

Variable	PCC			PCC-A		
	OR	95% CI	<i>p</i>	OR	95% CI	<i>p</i>
UCLP (n=62)	REF			REF		
BCLP (n=17)	-3.488	-33.340-6.363	.483	-3.031	-12.945-6.882	.545
SHCP (n=21)	-3.389	-13.769-5.993	.436	-4.568	-14.511-5.374	.364
Cleft width	-0.676	-1.172- -0.179	<.01	-0.604	-1.104- -0.105	<.05

UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, OR = odds ratio, CI = confidence interval

There was no significant difference for CSCs between cleft types (BCLP 65%, UCLP 58%, SHCP 57%, $\chi^2(2) p = .869$). Oral and non-oral articulation errors were more common in individuals with BCLP (59% and 24%, respectively) compared to those with UCLP (52% and 15%, respectively) and SHCP (48% and 14%, respectively).

No significant differences between cleft types were found for DSCs (BCLP 65%, SHCP 57%, UCLP 50%, $\chi^2(2) p = .531$). When analyzing subtypes of DSCs, velar fronting was rare but most common in individuals with SHCP (10%) compared to those with BCLP and UCLP (0% and 3%, respectively).

Stopping was marginally more common in individuals with SHCP (14%) compared to those with BCLP and UCLP (12% and 11%, respectively), as was voicing errors (14%) compared to those with BCLP and UCLP (12% and 10% respectively).

Having both CSCs and DSCs was most common in the SHCP and BCLP groups (48% and 47%, respectively) compared to the UCLP group (27%). The most affected target sound was /s/ in all groups and was present in 65% of the BCLP group, 60% in the UCLP group, and 52% in the SHCP group. The s-errors mainly consisted of palatalization of /s/ or inter-dental realization of /s/.

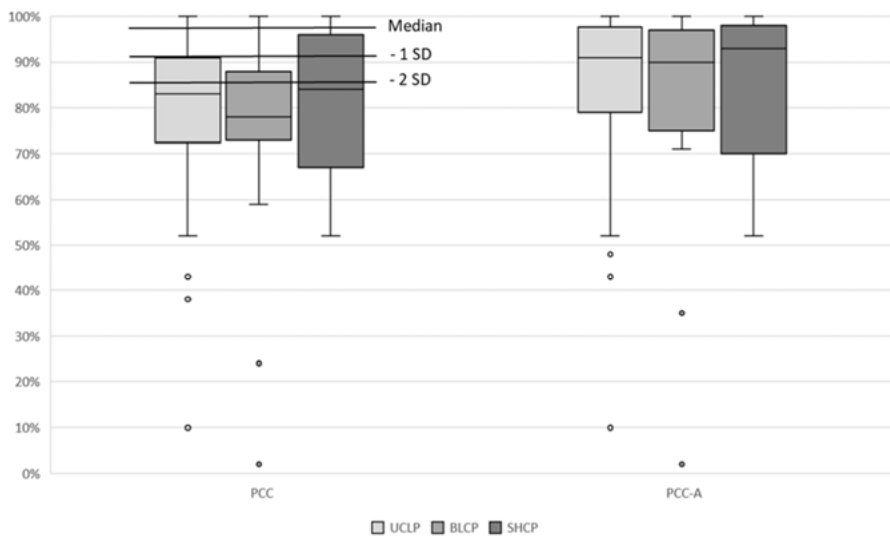


Figure 1. Comparison of median percent of correct consonant (PCC) and PCC adjusted for age (PCC-A) between cleft types. The norm PCC values for 5-year-old Swedish speakers without cleft palate at median, - 1 SD and - 2 SD, are indicated by horizontal lines. SD = standard deviation, UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SCHP = cleft in the soft and hard palate

Velopharyngeal function

Ordinal regression analysis showed that neither cleft type nor cleft width were significantly associated with VPC-Sum; see Table 8

Table 8. Ordinal regression analysis on how cleft type and cleft width were associated with VPC-Sum.

Variable	OR	VPC-Sum	
		95% CI	p
UCLP (n=62)	REF		
BCLP (n=17)	2.651	.941-7.470	.065
SHCP (n=21)	2.402	.790-7.301	.122
Cleft width	1.332	.755-2.351	.322

UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, SHCP = Cleft in the soft and hard palate, VPC = velopharyngeal competence, OR = odds ratio, CI = confidence interval.

Discussion

This study investigated the impact of cleft type and cleft width on the rate of secondary palatal surgery, articulation, and VPF. It also sought to examine any differences in types of articulation errors between cleft types.

No significant association was found between cleft type or width and the rate of secondary palatal surgery due to dehiscence, fistula, or VPI at five years of age. However, individuals with wider clefts and/or SHCP appeared more likely to require speech-improving surgery. In this study, the rate of speech-improving surgery was found to be 19% in the SHCP group, 18% in the BCLP group, and 10% in the UCLP group. These findings seem to contradict the results of some earlier studies that show a higher incidence of speech-improving surgery with more extensive clefts (18, 42) but are consistent with another study that reported speech-improving surgery rates of 18% for SHCP and BCLP groups and 9% for a UCLP group (17). The SHCP group in the whole cohort born between 2000 and 2015 constituted another 86 individuals, meeting the inclusion criteria of being treated at our unit since birth. Many of them were excluded because they had undergone 1-stage palatoplasty. The individuals included in the study likely had more severe and wider clefts and were possibly not considered suitable for 1-stage surgery. This could have resulted in poorer outcomes for the SHCP group than if those undergoing 1-stage palatoplasty had also been included in the study. This may explain the difference between our results and those showing that more extensive clefts more often require speech-improving surgery. Cleft width has been shown to be associated with the rate of secondary palatal surgery (4, 15). Though insignificant, this study also found a tendency of wider cleft more likely requiring speech-improving surgery.

The cleft type was not associated with PCC or PCC-A. All three groups performed substantially lower than typically developed children without cleft palate. The median PCC of just above or below 80% was more than two standard deviations (SD) below the 97% norm. A PCC of 91% equals - 1SD, which can be considered as age-appropriate. This was reached by 32% in the UCLP group, 29% in the SHCP group, and 18% in the BCLP group. In comparison, another study reported that 65% in the SHCP group, 33% in the UCLP group, and none in the BCLP group reached this level and that the difference between BCLP and SHCP and UCLP was statistically significant (18). When calculating PCC-A, the median improved to 91% in the UCLP group, 90% in the BCLP group, and 93% in the SHCP group, indicating that s-errors that are scored as correct in PCC-A account for the greater part of the articulation difficulties. The result of PCC-A differs somewhat from the statistically significant differences between comparable groups in an earlier study, with PCC-A at approximately 97% in the SHCP and UCLP groups and 86% in the BCLP group (17). The results for both PCC and PCC-A in this study's SHCP and UCLP groups were worse, but the results for the BCLP group were better than those reported in the abovementioned studies (17, 18).

No significant differences were observed between the groups when dividing articulation errors into CSCs and DSCs. It was more common with CSCs in the BCLP group, and 24% had more than one type of CSC compared to none in the SHCP group and 6% in the UCLP group. The most common type of CSC in all groups was retracted articulation to palatal or velar/uvular place. As in the Scandcleft studies (37, 38), differentiating between retracted articulation to palatal or velar/uvular place was not made in this study. However, this is differentiated when using the Cleft Audit Protocol for Speech – Augmented CAPS-A (43, 44). In order to compare, we analyzed further and found retracting to palatal place to be present in 38% of the SHCP group, 47% in the BCLP group, and 45% in the UCLP group. Retracting to velar/uvular place was present in 5% of the individuals with SHCP, 6% in those with BCLP, and 10% in those with UCLP. In comparison, the percentage of individuals who exhibited backing to palatal place was 5.4% in the SHCP group, 31.9% in the BCLP group, and 14% in the UCLP group. Similarly, the percentage of individuals who exhibited retracting to velar/uvular place was 3.6% in those with SHCP, 17.6% in those with BCLP, and 9.7% in those with UCLP (2). The results about the proportion of children having a specific articulation error are not entirely comparable, as our cutoff of three or more occurrences signified an error, and Butterworth et al. (2) used a cutoff of one occurrence. However, the palatal or velar/uvular realization distribution in the BCLP groups differs. In our BCLP group, substantially more children exhibited retracted articulation to palatal place than retracted articulation to velar/uvular place, while the proportions did not differ as much in Butterworth et al. (2). The differences noted between the studies may be explained by difficulties in differentiating between palatal and velar/uvular articulation.

Individuals with BCLP more commonly had DSCs, but it was more common in the SHCP group to have more than one type of DSC. 19% in the SHCP group had more than one type of DSC compared to only 6% in the BCLP group and 8 % in the UCLP group. Individuals with UCLP were less likely to have both CSCs and DSCs. Although the number of individuals having different types of CSC and/or DSC differed between cleft types, there were no differences regarding the type of articulation error. In all groups, /s/ was the most affected target sound and was present in 65% of the total group.

Overall, individuals with BCLP had worse outcomes at the group level, which aligns with previous studies (2, 10, 16-20). However, they did not differ significantly from the two other groups in the present study. Cleft width, on the other hand, was significantly associated with PCC and PCC-A. Cleft width is related to cleft type (45), meaning wider clefts are more common in more extensive clefts. However, this does not translate into clefts involving more structures having less favorable outcomes than those involving fewer structures. Indeed, our results indicate that a narrow but extensive cleft may have better outcomes than a wide but less extensive cleft. Therefore, we must consider the cleft width to make more accurate predictions at an individual level.

One limitation of this study is the small sample size. Larger groups may have yielded other results, and the results should be interpreted with caution. Another limitation of this study is the lack of hearing status. The impact of hearing impairment on speech outcomes varies among studies, but its impact cannot be ruled out. A significant correlation between the history of articulation and hearing difficulties has been shown (10, 46), and hearing levels at 18 and 36 months correlated to PCC at three years of age (47). Contrary to these findings, research has shown that hearing was not linked to speech outcomes at three (36) or five years old (19).

Conclusions

In this study, cleft width predicted PCC and PCC-A, and even though it did not significantly impact the rate of secondary surgery or VPF, cleft width should be considered when searching for factors that may affect different outcome measurements. The cleft type was not significantly associated with any outcome measurements and may not be regarded as a reliable predictor.

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