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Speech outcome in children born with cleft lip and palate
treated with one-stage palate repair:
aspects of function and environment

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Hand brushed Japanese ideogram

Myō ≈ curious, fantastic, irregular, obvious, rare,
remarkable, strange, wonderful

SPEECH OUTCOME IN CHILDREN BORN WITH CLEFT LIP AND PALATE: ASPECTS OF FUNCTION AND ENVIRONMENT

THESIS FOR DOCTORAL DEGREE (Ph.D.)

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ABSTRACT

Introduction and aims: This project focused on speech outcomes at 5 and 10 years of age after two different surgical protocols and on peer perceptions of speech in children born with cleft palate. The aim was to gain knowledge about speech outcome in children born with unilateral cleft lip and palate (UCLP) or cleft palate only (CPO), treated with one-stage palatoplasty at approximately 12 months of age, according to two different surgical protocols: minimal incision technique (MIT) and minimal incision technique with muscle reconstruction (MITmr). In addition, the aims included to study how peers not familiar with the speaker describe cleft related speech impairment in their own words and the possible consequences that it might have in a social context.

Materials and methods: A total of 217 children born with a cleft palate were included in this project: 69 children with UCLP and 148 children with CPO. In the CPO group, 98 had a cleft in both the hard and soft palate (CPH) and 50 in the soft palate only (CPS). The children were born in the Stockholm region in 1987–2004. In addition, 37 children born without a cleft participated: 18 children as a reference group at 5 years of age and 19 children in focus groups at 10 years of age. Speech outcomes were assessed from audio recordings by external expert speech and language pathologists. Intra- and inter-rater reliability were determined. Three focus groups of peers listened to and described speech samples with different types of cleft speech characteristics as well as normal speech.

Results: The children with different cleft types presented a high degree of hypernasality and audible nasal air leakage at 5 years of age, especially in the group operated on according to the MIT protocol. A significant decrease at age 10 was found, except for audible nasal air leakage in children with CPO. With the MIT protocol 43% of the children with UCLP, 42 % with CPH and 4% with CPS had received a velopharyngeal flap. The prevalence of articulation errors, such as retracted oral articulation and glottal articulation, decreased with age. The CPO group showed fewer articulatory errors than the UCLP group. Peers identified normal speech and more pronounced signs of nasality and easily detected and reacted upon even single occurrences of articulatory difficulties and commented on possible reasons why and how these errors could influence communication.

Conclusions: The prevalences of nasality and articulation errors were similar to what is commonly reported at 5 and 10 years of age in children born with cleft lip and palate. Nasality and articulation improved with age, except for nasal air leakage in the CPO group, indicating that more velopharyngeal flaps might be required at a later age. In the CPO group, were velopharyngeal flaps common in children with CPH treated with MIT. Velopharyngeal flaps were also common in the UCLP group treated with MIT. Peers detected and commented on moderate to severe nasality and articulatory difficulties including minor errors. This should be considered when planning intervention.

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SAMMANFATTNING

Introduktion och syfte: Fokus i detta projekt var dels att undersöka talet vid 5 och 10 år efter två olika operationsmetoder av gommen hos barn födda med läpp-käk-gomspalt, dels att få en uppfattning om hur jämnåriga uppfattar talet. Syftet var att erhålla utökad kunskap om talresultatet efter den kirurgiska behandlingen av gomspalten i ett steg vid ca 12 månaders ålder hos barn födda med enkelsidig läpp-, käk- och gomspalt samt isolerad gomspalt. Metoden för gomoperation var antingen “minimal incision technique” (MIT) eller “minimal incision technique med muskelrekonstruktion” (MITmr). Ytterligare ett syfte var att undersöka hur jämnåriga, med egna ord, beskriver talet hos 10-åringar som är opererade för gomspalt och har olika typer av talavvikelser.

Material och metoder: Totalt 217 barn födda med gomspalt inkluderades i detta projekt: 69 med enkelsidig läpp-käk-gomspalt och 148 med isolerad gomspalt. I den senare gruppen hade 98 barn en spalt i den hårda och mjuka gommen och 50 barn en spalt i enbart den mjuka gommen. Barnen var födda i Stockholmsregionen mellan år 1987–2004. Ytterligare 37 barn utan gomspalt deltog i projektet: 18 barn utgjorde en referensgrupp i en delstudie och 19 tioåringar medverkade i fokusgrupper. Audioinspelningar användes för standardiserad lyssnarbedömning av externa logoped, experter i ämnet. Intra- och interbedömarreliabilitet beräknades. Tre fokusgrupper med jämnåriga lyssnade på och beskrev talexempel med olika spaltrelaterade talavvikelser, men också med normalt tal.

Resultat: En hög förekomst av hypernasalitet och hörbart nasalt luftläckage förekom vid 5 år ålder, framför allt i gruppen opererad med MIT. Resultaten visade en signifikant minskning vid 10 års ålder, förutom hos gruppen med isolerad gomspalt, hos vilka hörbart nasalt luftläckage ökade. I gruppen barn med enkelsidig läpp-käk-gomspalt, opererade med MIT, hade 43% av barnen erhållit talförbättrande kirurgi (velofarynxlambå). I gruppen med isolerad gomspalt hade 42% av barnen med spalt i hårda och mjuka gommen och 4% av barnen med spalt i mjuka gommen erhållit en sådan tilläggsoperation. Förekomsten av artikulationsavvikelser, t.ex tillbakaflyttad oral artikulation till velar eller glottal artikulation, minskade med åldern. Gruppen med isolerad gomspalt hade färre artikulatoriska avvikelser än gruppen med enkelsidig läpp-käk-gomspalt. De jämnåriga identifierade normalt tal och reagerade på mer uttalade tecken på nasalitet men även på enstaka artikulatoriska avvikelser. De spekulerade i orsakerna och kommenterade också hur dessa skulle kunna påverka barnens kommunikativa situation.

Slutsatser: Förekomsten av nasalitet och artikulatoriska avvikelser var i nivå med vad som ofta rapporteras vid 5 och 10 års ålder hos barn födda med läpp-käk-gomspalt. Hörbart nasalt luftläckage ökade dock i gruppen med isolerad gomspalt, vilket kan tyda på ett behov av ytterligare talförbättrande kirurgi. Hos barn med isolerad gomspalt var velofarynxlambå vanligast hos de med spalt i hårda och mjuka gommen, opererade med MIT. Velofarynxlambå var också vanligt hos barn födda med enkelsidig läpp-käk-gomspalt opererade med MIT. Jämnåriga 10-åringar förefaller notera mer framträdande nasalitet, medan även lättare, enstaka artikulationsavvikelser uppmärksammades. Detta bör beaktas vid planering av intervention.

LIST OF SCIENTIFIC PAPERS

- I. **Nyberg J**, Raud Westberg L, Neovius E, Larson O, Henningsson G.
Speech results after one-stage palatoplasty with or without muscle reconstruction. *The Cleft Palate-Craniofacial Journal* 2010, 47(1): 92-103.
- II. **Nyberg J**, Peterson P, Lohmander A.
Speech outcomes at age 5 and 10 years in unilateral cleft lip and palate after one-stage palatal repair with minimal incision technique – A longitudinal perspective. *International Journal of Pediatric Otorhinolaryngology* 2014, 78: 1662-1670.
- III. **Nyberg J**, Neovius E, Lohmander A.
Speech outcomes at 5 and 10 years of age after one-stage palatal repair with muscle reconstruction in children born with isolated cleft palate. Submitted manuscript.
- IV. **Nyberg J**, Havstam C.
Speech in 10-year-olds born with cleft lip and palate: what do peers say?
The Cleft Palate-Craniofacial Journal 2016, 53(5): 516-526.

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1 INTRODUCTION

1.1 CLEFT LIP AND PALATE

Cleft lip and palate is the most common congenital craniofacial malformation, which is due to a failure of fusion of the structures forming the different parts of the lip and palate during the 5th to 12th week of pregnancy. The incidence in Sweden is about 2/1000 births, which results in approximately 150–200 births every year with some type of cleft (Hagberg et al., 1998; Mossey, 2007). The different cleft types are usually divided into unilateral or bilateral clefts of the lip, alveolus and palate, and isolated cleft palate. The extent of the cleft palate may vary from a partial cleft in the soft palate to a complete cleft of both the hard and soft palate. About 35% of the children are born with a unilateral cleft lip and palate (Figure 1a) and another 40% are born with an isolated cleft palate (Hagberg et al., 1998). The isolated cleft palate can include both the hard and the soft palate (Figure 1b) or the soft palate only (Figure 1c). The incidence of associated anomalies or syndromes is higher among children with a cleft palate only than in other cleft types (Watkins et al., 2014). The figure varies between 22% to 70%, probably due to diverse definitions (Shprintzen et al., 1985; Milerad et al., 1997).

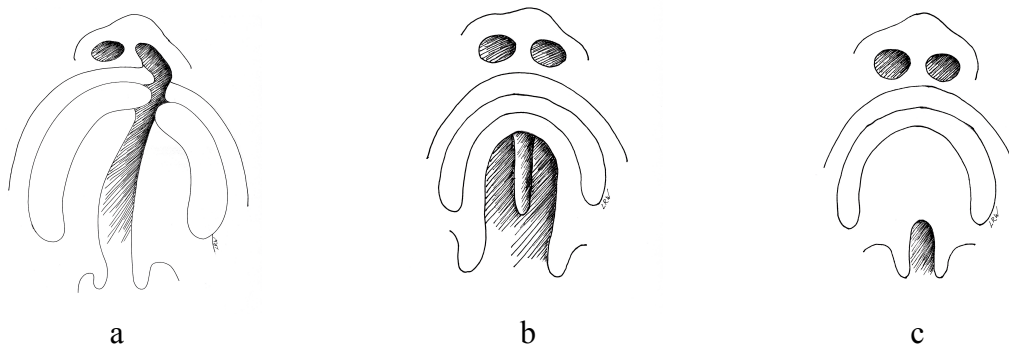


Figure 1. (a) Unilateral cleft lip and palate. (b) Isolated cleft palate with a cleft in both the hard and soft palate. (c) Isolated cleft palate with a cleft in the soft palate only. Illustrations by Liisi Raud Westberg.

1.2 CLINICAL MANAGEMENT OF CHILDREN WITH CLEFT LIP AND PALATE

Depending on the type and the extension of the cleft, different functions might be affected, such as eating, babbling, speech development, ear function/hearing, dentition and facial appearance. To achieve optimal development of these structures and functions, the cleft is surgically closed. The condition after primary repair of the cleft palate may require secondary speech improving surgery and/or speech therapy. Orthodontic treatment is almost always needed. In addition, children born with a cleft palate are more affected by hearing impairment, due to otitis media with effusion. A multidisciplinary team including plastic surgeons, orthodontists, speech and language pathologists, otorhinolaryngologists, audiologists, psychologists and nurses collaborate and provide the treatment.

The cleft palate teams in Sweden follow the national guidelines and recommendations regarding care for a child born with cleft lip and palate. In Stockholm, the family meets a team member at the maternity ward and receives the initial information. Thereafter, the child's treatment is planned, following the national routines for documentation, but according to the regional surgical protocol. Thus, the surgical protocols vary at the different cleft palate centres, whereas the documentation and assessment of speech and maxillary growth and occlusion follow standardised care procedures. In order to ensure the quality of care in a systematic way, data from surgical protocols, orthodontic treatment and speech outcome are registered in the Swedish National Quality Register from 5 years of age, with additional registrations at 10, 16 and 19 years of age (www.clpregister.se/2-nationella-varldprogrammet-for-lkg).

1.3 INTERNATIONAL CLASSIFICATION OF FUNCTIONING, DISABILITY AND HEALTH

The International Classification of Functioning, Disability and Health (ICF; WHO 2001), is a tool to describe and classify human functioning and health and focuses more on the impact rather than the cause of a disorder. The overall aim of the classification system is to provide a structure and a standardised language to describe both the functional ability and disability in relation to health. It consists of two main parts: 1. Function and disability and 2. Contextual factors. The first component relates to body functions and structures as well as activities and degree of participation. Contextual factors relate to environmental and

personal factors. The Children and Youth Version (ICF-CY, 2007) was derived from ICF, specifically designed to take the changing nature of children's health and development into account (McGormack et al., 2010; Neumann & Romonath, 2012). Cleft speech problems do have a physical basis, but for a wider, more holistic view of the impact of cleft speech impairment, the ICF framework might be helpful to understand the impact on the affected child. Communicative participation can be studied in different ways — both from an individual point of view and also to include the childrens environment, and to identify possible barriers and facilitators in their social context (Havstam & Lohmander, 2011). In order to identify speech impairment, the disability is often seen as a problem of the affected person, which requires medical care provided by professionals (World Health Organisation, 2001). It is unclear whether this medical approach reflects the communication experience of children with speech difficulties and their communication partners (McGormack et al., 2010).

1.4 ASPECTS OF STRUCTURE AND FUNCTION

1.4.1 Anatomy of the palate and the velopharynx

The palate comprises the hard and soft palate. The bony hard palate is anteriorly placed and merges posteriorly to the mobile muscular soft palate (the velum). The soft palate extends from the posterior border of the hard palate and four pairs of muscles form the soft palate: the tensor veli palatini tenses and the levator veli palatini raises the velum, the palatoglossus and palatopharyngeus depresses the velum. The palate develops between the 4th and 12th week of pregnancy. At several stages this development can be disrupted. The midline in the hard palate marks the fusion of the two palatal shelves, starting from the anterior part moving back, like a zipper. Figure 2 illustrates the cleft muscular anatomy compared to the anatomy of a normal palate.

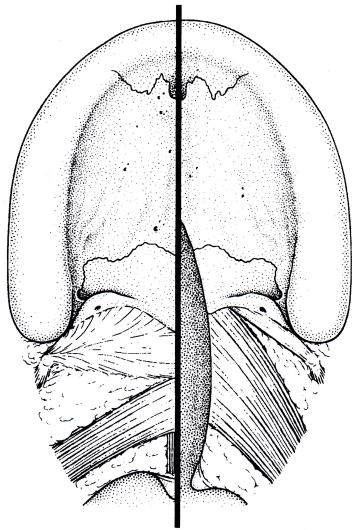


Figure 2. A diagram of the cleft muscular anatomy, with the anatomy of the cleft on the right side and the normal anatomy on the left side. The right side illustrates the abnormal triangular insertion of the tensor palatini and the anterior insertion of the levator muscle. The left side, with the normal anatomy illustrates the tensor tendon/palatal aponeurosis thinning posteriorly and occupying the anterior third of the velum and the levator in the middle third. Published with permission from Mr B Sommerlad and John Wiley & Sons Ltd.

Fundamental for normal speech production, is that the passage between the oral and nasal cavity can be separated using the velopharyngeal structures: the soft palate (velum) and the pharyngeal walls (pharynx). The aim of palatal surgery is to enable a division between the cavities in order to achieve a fully functional velopharyngeal system (Figure 3).

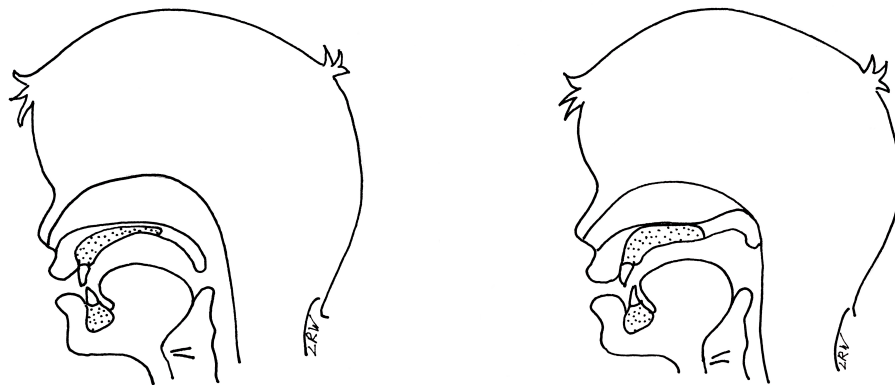


Figure 3. Velofarynx during breathing to the left and with sufficient velopharyngeal closure to the right. Illustrations by Liisi Raud Westberg.

An insufficient velopharyngeal closing mechanism might occur after palatal surgery as a result of a short palate, a disproportion between the palatal length and the depth in the pharynx, or reduced mobility of the velum and pharynx (Figure 4).

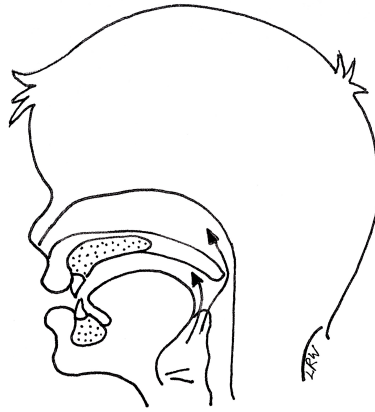


Figure 4. Insufficient velopharyngeal closing mechanism. Illustration by Liisi Raud Westberg.

Other reasons for an oro–nasal coupling might be a fistula in the palate due to a rupture after surgery, or if a residual cleft is left in the alveolar ridge.

1.4.2 Surgical protocols for palatal surgery

The goal of palatal surgery is to provide the best results regarding both adequate midface growth and normal speech outcomes. Over the years, palatal surgery has tended to be performed at younger ages to promote beneficial speech development. The most common practice is to perform one-stage palate repair at around 12–15 months of age. With this method both the hard and soft palate are closed in one session (Rohrich et al, 2000). In a two-stage protocol, soft palate repair is usually performed first at around 6 months of age simultaneously with primary lip surgery and correction of the nasal cartilage, followed by hard palate closure performed at a later age, commonly from 2 years of age. The purpose of the delayed hard palate closure is to minimise maxillary growth impairment (Friede, 2009). Furthermore, the surgical techniques in practice vary. A minimal incision palatoplasty to minimise scarring and the risk for midface disturbance was described by Mendoza et al. (1994) and later by Kriens (1997). A technique with more radical muscle reconstruction of the soft palate is often recommended today, where studies have claimed better speech results (Sommerlad, 2003; Andrades et al., 2008; Oyama et al., 2016). In children born with UCLP the residual cleft in the alveolar ridge is closed by a bone graft from the iliac crest at

approximately 7-12 years of age, before the permanent teeth erupt. In Sweden, both one and two-stage surgical protocols are performed.

If the primary palatal repair results in adequate physical structures for the establishment of oral articulation, oral direction of the vocal airstream and a normal resonance, one surgical operation is usually enough. However, if the surgical treatment is unsuccessful, resulting in a short palate with accompanying velopharyngeal impairment (VPI), affecting resonance and nasality negatively, secondary palatal surgery is warranted. One way to prolong the soft palate and improve the velopharyngeal function (VPF) is to perform a velopharyngeal flap. With this method a superiorly based soft-tissue bridge is dissected from the pharyngeal wall and inserted into an incision created on the nasal surface of the soft palate (Riski, 2011). The velopharyngeal flap helps the velum to elevate towards the nasal cavity and to activate the pharyngeal walls with the purpose of reducing velopharyngeal insufficiency (Figure 5).

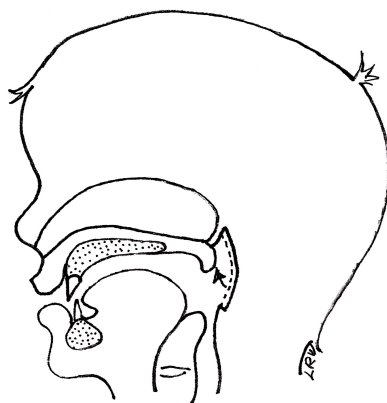


Figure 5. Velopharyngeal flap. Illustration by Liisi Raud Westberg.

1.4.3 Cleft speech characteristics

The features of speech production associated with a cleft palate can be related to the anatomical and functional constraints that the cleft imposes on the speech mechanism. In children born with a cleft palate, primary resonance and articulation may be impaired. Nasal resonance is present in normal voice production on nasal consonants. Deviances related to the abnormal coupling of the oral and nasal cavities concerns resonance (hyper and hyponasality) and also aerodynamics (nasal air leakage and weak pressure

consonants) (Grunwell & Sell, 2001). The term nasal air leakage describes inappropriate emission of air through the nasal cavity or friction in the velopharyngeal area during the articulatory production of high-pressure sounds (Figure 4). These symptoms can also be described as passive cleft speech characteristics, which are related to the inability to separate the two cavities (Hutters & Brøndsted, 1987). The other main category of cleft speech characteristics often named compensatory articulation, is related to the reduced ability to produce the speech sounds in the correct place of articulation due to the anatomical deviances. As a strategy to compensate for the inability to produce intra-oral pressure needed for high-pressure consonants, i.e. oral stops such as /t/, or fricatives such as /s/, are produced behind the origin for the oro-nasal coupling (Henningsson & Isberg, 1990). This is also described as active speech characteristics, since the strategy is to find the best place of articulation in order to produce a distinct, high-pressure consonant (Harding & Grunwell, 1998). For example, the influence of an oro-nasal opening such as a residual cleft in the alveolus or hard palate may result in retracted oral articulation, meaning that consonants normally produced anteriorly, such as the dental oral stop /t/ (Figure 6a) is produced at the velar position, where it is possible to obtain enough intra-oral pressure for oral stop production. The /t/ will thus be realised as [k] (Figure 6b). Furthermore, the production of /s/ might be influenced by dental or occlusionl changes. Glottal articulation refers to consonants being produced at the vocal cord level (glottis). The high pressure consonants can not be produced orally due to an insufficient velopharyngeal function and are consequently produced as non-oral glottal consonants (Figure 6c).

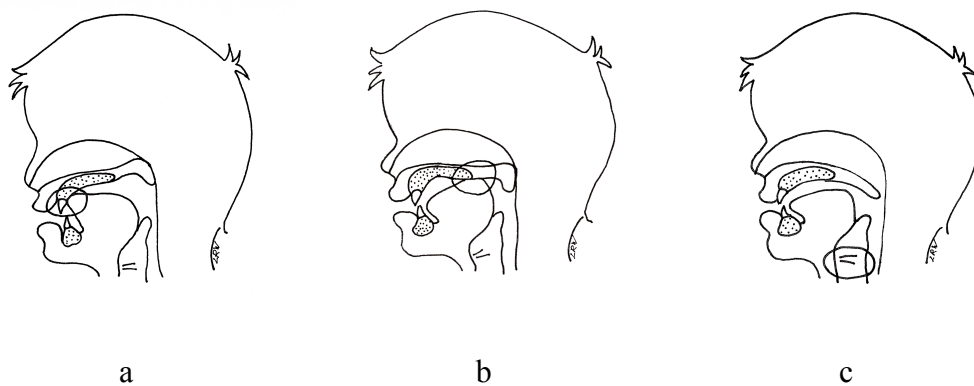


Figure 6. (a) Correct place of dental/alveolar articulation. (b) Articulation retracted to velar place of articulation. (c) Articulation retracted to vocal cord (glottal) level. Illustrations by Liisi Raud Westberg.

1.4.4 Speech assessment

Perceptual speech evaluation is the basis for speech assessment. Blinded analysis of speech data by independent specialist SLPs is the gold standard methodological approach when reporting audit and research outcomes (Lohmander & Olsson, 2004; Sell, 2005). Today the routines for speech assessments start with documentation and analysis of the early speech development, i.e. babbling and consonant inventory at 18 months of age. The Swedish Articulation and Nasality Test, SVANTE, (Lohmander et al., 2015) is the standardised test for speech assessment used by all Swedish speech and language pathologists in the cleft care teams. Articulation and nasality are evaluated at 3, 5, 7, 10, 16 and 19 years of age. Normative data in Swedish are available at all ages (Lohmander et al., 2016). In the 1990s standardised speech material was not yet available, but in Stockholm, as well as in the other cleft care centres, the same sentences including specially selected target consonants, including both high and low pressure consonants, similar to the ones in SVANTE were elicited by repetition from 5 years of age. Sentence repetition is considered as reliable and valid speech material (Klintö et al., 2011). Over the past 20 years the equipment for audio recording of speech has progressed rapidly, from the analogue Revox, to the digital DAT and H4nex Handy recorder. The audio recordings from the latter are stored directly in the child's medical records. In addition, high quality microphones are able to register more subtle speech deviances.

1.4.5 Cleft palate and speech outcomes

The ages when the children are about to enter school and adolescence are the most commonly reported outcome ages in the literature regarding speech outcomes related to cleft palate. According to a review of studies where the assessments were performed from audio/video recordings, 60% to 70% could be expected to have good speech at 4 to 5 years of age. The corresponding figure at around 10 years of age was around 80% (Lohmander, 2011). In a national prospective study in the United Kingdom, Sell et al. (2001) reported on speech outcomes at 5 years of age in over 200 children born with UCLP, treated with varied methods for surgical repair of the palate. The results showed that 34% of the children had at least one serious error in consonant production, 29% some degree of hypernasality and less than 20% had entirely normal intelligibility. Poorer results regarding hypernasality in the same cleft type was presented by Pigott et al. (2002) in their comparison of speech outcomes at 5 years of age in 66 children after three different

surgical methods for one-stage palatal repair. They found that around 45% of the 5-year-olds had hypernasality, with no significant difference related to the surgical method. The occurrence of cleft-related articulation errors was also high but varied between the surgical methods (32%–78%). Pulkkinen et al. (2001) evaluated speech in 30 children with UCLP at different ages including 6 years of age and showed that hypernasality was present in 41.5%. Compensatory articulations were present in 12% of the children at the same age. Timmons et al. (2001) reported better speech outcomes regarding hypernasality in 17 children with UCLP, but anterior oral articulation errors were found in 53% of the children.

It has been suggested that the cleft extent has an impact on speech outcome in children born with CPO – that is, the larger the extent of the cleft, the higher the impact on both velopharyngeal function (Brunnegård & Lohmander, 2007; Nyberg et al., 2010; Bicknell et al., 2002) and articulation (Persson et al., 2002). The influence of the cleft extent on articulation reported in earlier studies (Jakobsson et al., 1990; Lohmander-Agerskov et al., 1993), was confirmed in the study by Persson et al. (2006), who showed a higher prevalence of retracted oral articulation in children with CPH than in children with CPS.

In order to gain reference data and to provide a standard for benchmarking, Britton et al. (2014) published a large, multicentre cohort study including 1110 5-year-olds with different clefts involving the palate. In the study, 48% had normal speech and 60% had no serious cleft-related articulation errors. These results varied at different centres but confirmed their standard that 50% of the children should have normal speech, without any cleft-related speech errors, at 5 years of age. In the Scandcleft project, where three different surgical protocols for primary palatal closure were compared to a fourth common in three parallel randomised trials, 35% to approximately 60% had velopharyngeal competence and no hypernasality at 5 years of age with no significant differences between the different arms in the trials (Lohmander et al., 2016). In addition, around 20% consonants errors were reported in the trials (Willadsen et al., 2016). Sell et al. (2001) investigated a large group of 218 children with UCLP at 12 years of age and found that 19% had speech that was different enough to provoke comments. In contrast, speech has been reported to improve for most children at around that age (Park et al., 2000; Lohmander et al., 2006, Lohmander et al., 2012). In these studies of children treated according to different surgical procedures, the majority had normal speech by the age of 10 to 12 years. However, two small-sample cross-sectional studies of speech at 10 years of

age in children born with UCLP reported poorer outcomes on hypernasality and also on articulation: one after a two-stage palatal surgical protocol and one after a one-stage procedure. The results showed that between 34% and 46% of the 10-years-old children in these two studies had speech impairments (Brunnegård & Lohmander, 2007; Hortis-Dzierzbicka et al., 2012).

Since the treatment for excessive hypernasality is velopharyngeal surgery, the need for this type of secondary surgery is sometimes used as an indicator of the success rate regarding the primary palatal operation. The frequency of velopharyngeal flaps in children with UCLP in the studies above varied between 10% and 20% with no clear difference related to the performed primary palatal surgical procedure. In the Scandcleft project, however, the significantly highest pharyngeal flap rate (23%) was reported in one of the trials (Lohmander et al., 2016). Timmons et al. (2001) reported that velopharyngeal impairments measured in terms of a high velopharyngeal flap rate were significantly more common in the CPO group (37%) compared to the UCLP group (none).

1.4.6 Cleft palate and speech therapy

The assumption that the number of children with speech errors considered to be in need of speech therapy will decrease with improved surgical treatment programmes has until now not been proven correct, and the empirical evidence regarding the impact of speech therapy for children with a cleft palate is limited in both quantity and design (Peterson-Falzone et al., 2010). In a recent systematic review, Besell et al. (2013) found no evidence to support any specific speech therapy intervention. However, several studies have revealed that many children receive considerable speech intervention, either a combination of surgery and therapy or one of them, depending on the speech deviances. For example Sell et al. (2001) reported that approximately two-thirds of both 5- and 12-year-olds with UCLP had undergone speech therapy due to articulation errors, and in 50–60% the therapy lasted for more than one year. These figures are equal to those presented by Brunnegård and Lohmander, (2007) and Pulkkinen et al. (2001) who reported that 67% and 57%, respectively, had received speech therapy, although no details about the content in the therapy were provided. A high percentage of children receiving speech therapy was reported in the Scandcleft project as well, however with large variations. Thus, whereas some children received no sessions some had received over 100 session until age 5. It was

concluded that the effectiveness in speech therapy interventions needs to be evaluated (Lohmander et al., 2016; Willadsen et al., 2016).

1.4.7 Cleft palate and otological status

It is well known that infants and young children with a cleft palate demonstrate an abnormal middle ear status and that they are at risk for recurrent otitis media with effusion (OME), (e.g. Flynn et al., 2009). Although the prevalence declines with age it often remains at pre-school ages and beyond. For example, 63% at age 5 and 33% at age 10 years has been reported to have abnormal middle ear status (Flynn & Lohmander, 2014). OME is associated with a conductive hearing loss between 30-45 dB (Sheahan et al., 2003). Mild to moderate hearing loss is considered a potential threat to speech and language development and routine follow-up of ear status and hearing is recommended (Peterson-Falzone et al., 2010). The association with speech and language development in children with cleft palate is still unclear due to lack of studies in the area. No relationship between otitis media and delayed speech and language development has been found in typically developing children (Zumach et al., 2010).

1.5 ENVIRONMENTAL ASPECTS

1.5.1 Perception of speech

A listeners attitudes and judgments about a person's age, health, social and educational status are often based on the person's speech characteristics (Allard & Williams, 2008). Negative stereotyping, due to communication disorders, is of high importance to society, since it may cause psychological, educational, and employment disadvantages (Lee et al., 2016). Also in children negative attitudes toward peers with speech impairments have been observed and subjective perceptions of speech have been related to social acceptance (Blood & Hyman, 1977; Watterson et al., 2013).

Peer acceptance has been shown to be crucial for the development of social communication abilities and also for feeling socially included (Feragen et al., 2009). Frederickson et al. (2006) found that there appeared to be a relationship between speech production skills and conversational skills, suggesting that poor speech may be impacting communication in

children with a cleft palate. The speech of children with communication disorders has also been found to influence listeners' perceptions of the speakers' non-speech characteristics. Studies of different types of speech impairment, such as dysarthric speech and stuttering, indicate that others draw negative inferences about the personality of the person speaking (Lass et al., 1991; Witt et al., 1997; Franck et al., 2003). Further, speech disorders may affect teacher's expectations of student achievement negatively (Overby et al., 2007). Research including children with both speech impairments and facial disfigurements indicates that these children are at risk of being underestimated by their teachers with regard to their intelligence (Millard & Richman, 2001) and that teachers view children born with a cleft lip and palate as more inhibited in the classroom compared with what their parents observe at home (Richman, 1978). If children with a speech impairment are met with lower expectations by teachers and peers in the classroom, it might have a negative impact on their achievements (Feragen et al., 2015). Teasing has been associated with a child's psychosocial functioning and if prevalent at 10 years and thereafter it might impact depressive symptoms (Feragen et al., 2009). In a study by Semb et al. (2005), 74% of the children reported having being teased. The focus of teasing was appearance of the nose and lip, followed by speech.

Similarities between perceptual judgment by SLPs and untrained listeners have been reported (Brunnegård et al., 2009), while suggestions that ratings done by professionals do not necessarily reflect the attitudes of the community (Witt et al., 1996; Dagenais et al., 2006) have been presented. In order to add real-life significance to clinical speech assessments, Kuehn and Moller (2000) advocated that outcome measures for speech also should include judgement from the environment. Witt (1996) concluded that peer group evaluations of cleft palate speech are more relevant to the patients themselves. Another factor of importance is a person's own satisfaction with treatment. Several researchers have reported a relationship between self-reported speech problems and self-reported teasing in young individuals with some type of cleft palate (Hunt et al., 2006; Noor & Musa, 2007; Havstam et al., 2011). Havstam et al. (2011) investigated communication attitudes in 10-year-olds with a cleft lip and palate using the Swedish version of the Communication Attitude Test (CAT-S) and compared the answers with parents' answers about satisfaction with their children's speech. They found that there were significant correlations with their opinions of communicative competence. Although there seems to be a relationship between speech performance and self-reported communication attitudes in 10-year-olds born with some type of cleft palate, not all children with impaired speech develop negative communication attitudes (Havstam et al., 2011). In one study, 35% of the children with clefts reported that they often/very often had to

repeat a verbal message due to reduced speech intelligibility, but nevertheless 91% were satisfied with their speech (Van Lierde et al., 2012). Subjective perceptions of speech have also been related to social acceptance (Watterson et al., 2013).

1.5.2 Qualitative perspectives in cleft palate research

Qualitative research is concerned with making sense of people's beliefs and behaviours (Nelson, 2009). In order to widen the perspective in the area of craniofacial research, qualitative methods are becoming more frequent. Earlier studies in the field have mostly concerned different craniofacial conditions, some including a cleft, and they have focused on the experience of living and coping with appearance-related challenges (Marcusson et al., 2002; Roberts & Shute, 2010; Pope et al., 2016). However, it might be difficult to tease out issues of specific interest to children with CLP (Sharif et al., 2013). Furthermore, the current research findings need to be complemented with patient-focused information that may encourage the cleft care teams to revise and improve their treatment methods and also to bridge the gap between research and practice. In the field of cleft lip and palate, qualitative methods with their flexible, socially responsive approach might offer important ways to facilitate the involvement of children in issues related to their care and to highlight the questions important in real life (Nelson, 2009; Havstam, 2010; Sharif et al., 2013).

1.5.3 Qualitative content analysis and focus groups

Qualitative content analysis is one of several qualitative methods, often used when the data collection is based on interviews, especially if the study deals with an unknown topic (Patton, 2002; Hsieh & Shannon, 2005; Krippendorff, 2013). Content analysis focuses on trying to describe variations of phenomena, and is recommended for interpreting data from focus groups (Elo & Kyngäs, 2008). The analysis aims to create categories that are mutually exclusive and exhaustive and to discover themes. Focus groups offer an opportunity to discover how young people think about different issues in their environment (Krueger & Casey, 2009). Youth focus groups have been engaged in formulating questions for further inquiry in earlier research, for example in health care. Children and adolescents have provided information about food choices and eating behaviours, and physical exercise in order to form the basis for nutrition education and obesity intervention programmes. Different

health-related conditions have also been discussed, such as experiences with living with diabetes, asthma or a craniofacial difference (Peterson-Sweeney, 2005; Edwards et al., 2005).

1.6 RATIONALE FOR THE INCLUDED STUDIES

Children born with a cleft lip and palate are at risk for speech problems that may affect communication from an early age. The surgical protocols MIT and MITmr are well established today and have been performed by the Craniofacial Team in Stockholm since 1997. An evaluation of speech outcomes is important in order to determine the effectiveness of the treatment. An evaluation would preferably also include other aspects. Knowledge about the environment's reactions to cleft speech characteristics is still sparse – especially if and how peers perceive and describe speech in children born with cleft lip and palate and if they have opinions about the communicative ability and the possible consequences for social participation. One crucial question is whether peers notice all the traits that SLPs do and if some speech variables are more important for how peers perceive speech. It would be interesting to find out whether the treatment provided by the SLPs working with children born with CLP address the speech deviances that draw the most attention or cause the most problems for the individuals in their social context.

2 AIMS

2.1 GENERAL AIMS

The overall aims of the present project were:

- to provide knowledge about speech outcome and development in children born with UCLP or CPO, treated with one-stage palatoplasty at approximately 12 months of age.
- to investigate the speech results after the two different surgical protocols: MIT and MITmr.
- to explore how peers not familiar with the speakers describe cleft-related speech impairments.

2.2 SPECIFIC AIMS

The specific aims were to:

- examine if there was a difference in the occurrence of speech deviances in different cleft types (Study I, III) and compare it to a non-cleft reference group (Study I).
- study the prevalence of typical cleft speech errors at 5 and 10 years of age (Study II, III), related to the two different surgical methods.
- explore how 10-year-olds describe speech and the possible consequences speech impairments might have for the communicative participation in children with UCLP in their own words (Study IV).
- study whether peers register signs of VPI and articulation errors of different degrees and, if so, the terminology used (Study IV).

3 MATERIAL AND METHODS

3.1 PARTICIPANTS

A total of 217 children born with a cleft palate from the original cohort of 350 children participated in this project: 69 children with a unilateral cleft lip and palate (UCLP) and 148 children with a cleft palate only (CPO). In the CPO group, 98 had a cleft in both the hard and soft palate (CPH) and 50 in the soft palate only (CPS). The children were born in the Stockholm region between 1997 and 2004, and were treated by the Stockholm Craniofacial Team. In addition, 37 children born without a cleft participated, 18 children served as a reference group at 5 years of age and 19 children took part in the focus groups at 10 years of age. All children in the four studies were native Swedish speaking. In total, 133 children were excluded, 106 with CPO and 27 with UCLP. The reasons for exclusion were additional syndromes or malformations, developmental delay, unclear description of diagnosis, a different surgical technique used, international adoption, families had moved, lost to follow up, and poor quality of audio recordings. Consequently, a total of 217 children participated in the project (Table 1a).

Table 1a. Distribution of participants and number of exclusions in the four studies for the different cleft groups: CPO = cleft palate only, UCLP = unilateral cleft lip and palate, CPH cleft in the hard and soft palate, CPS = cleft in the soft palate only.

Cleft group	CPO		UCLP	Total
	254		96	350
Excluded	106		27	133
	CPH	CPS		
	98	50		
Total	148		69	217

A total number of 129 children were treated according to the surgical protocol, minimal incision technique (MIT) (Figure 7), 60 with CPO and 69 with UCLP. The corresponding figure for the protocol, minimal incision technique with muscle reconstruction (MITmr) (Figure 8), was 88 children with CPO (Table 1b).

Table 1b. Distribution of participants in the four studies for the different cleft groups and surgical techniques: CPO = cleft palate only, UCLP = unilateral cleft lip and palate, CPH cleft in the hard and soft palate, CPS = cleft in the soft palate only. MIT = minimal incision technique, MITmr = minimal incision technique with muscle reconstruction.

Surgical protocol	CPO		UCLP	Total
MIT	60		69	129
	CPH	CPS		
	36	24		
MITmr	88			88
Total	62	26	69	217

3.1.1 Study I

Two groups of children with non-syndromic CPO were included from a cohort of 133 children in this retrospective cross-sectional study of speech comprising children at 5 years of age born between 1987 – 1999. There were in total 86 children in the two groups: 53 with CPH and 33 with CPS, 37 boys and 49 girls. Forty-seven children were excluded for reasons given in section 3.1. Two surgical procedures for primary palatal had been performed. They were all carried out between 12 and 15 months of age. Sixty children were treated with MIT (Figure 7), 36 with CPH and 24 with CPS, and 26 children had a surgical treatment according to MITmr (Figure 8), 17 with CPH and nine with CPS. In the MIT group, 16 children (19%) received a velopharyngeal flap: 15 children with CPH and one child with CPS. In the MITmr group one child (with CPH) received a velopharyngeal flap. Data regarding speech therapy and hearing status was not obtained in this study. Eighteen children without a cleft served as a reference group, recruited from a preschool service in the Stockholm region. According to a parental questionnaire, the children's speech and language development was comparable to peers and the hearing was normal.

3.1.2 Study II

Study II, a retrospective longitudinal study of speech at 5 and 10 years of age, comprised 69 children with UCLP from a consecutive series of 96 children born between 1987 – 1997 (19 girls and 50 boys). Twenty-seven children were excluded for reasons given in section 3.1. All children were treated with the surgical protocol MIT (Figure 7) at a mean age of 13 months (SD 1.8). In addition, 30 children (43%) had received a velopharyngeal flap: 12 before the age of 5 and another 18 before the age of 10. At least 60% of the children had received speech therapy, and almost 20% had experienced over 30 therapy sessions. However, the type and exact amount could not be determined. Otological information at 5 years of age showed that 26% of the children had ventilation tubes at 5 years of age, and 36% had normal hearing status. However, the data was missing for 36% of the children and no audiological screening was performed at 10 years of age.

3.1.3 Study III

In this retrospective study with a longitudinal perspective, speech was investigated at 5 and 10 years of age in 88 children with non-syndromic CPO from a consecutive series of 147 children (62 with CPH and 26 with CPS, 43 boys and 45 girls), born between 1997 and 2004. Fifty-nine children were excluded for reasons given in section 3.1. Twenty-six children (17 with CPH and 9 with CPS) treated with MITmr (Figure 8) in study I were included, and all the children in the study group were treated with this same surgical protocol at a mean age of 13.6 (SD 4.6) months. Velopharyngeal flap surgery was performed on 12 children (14%) (10 with CPH and two with CPS), two before 5 and ten before 10 years of age. The type and the amount of speech therapy provided was difficult to determine from the hospital records. However, at least 40% of the children received different amounts of therapy regarding articulatory difficulties. Therapy might also have been administered by local SLPs or at schools. Regarding otological information, 41% of the children received ventilation tubes before 5 years of age and at 10 years of age, 7% had ventilation tubes; 44% had experienced a hearing test. However, the hearing data was not acquired at the same time as the speech assessment.

3.1.4 Study IV

In this descriptive study with a qualitative approach, nineteen 10-year-olds from the same school class were divided into three groups. Each group participated in a focus group interview. The parents answered a questionnaire regarding their child's speech development, hearing and previous contact with SLPs, and in addition whether the child had experience of a person with any kind of speech impairment. The purpose of the last question was to explore whether the children had discussed speech difficulties with their family. None of the children had a hearing disorder, was participating in ongoing speech therapy, or had recently spoken about speech difficulties with their relatives. The parents were kindly instructed not to inform their children about the purpose of the study.

3.2 SURGICAL PROTOCOLS

Two different surgical procedures for the palate were used and also compared in this project: the minimal incision technique (MIT) in study I and II (Figure 7) and the minimal incision technique with muscle reconstruction (MITmr) (Figure 8) in study I and III. Both methods are one-stage procedures, where the cleft in the hard and soft palate is closed simultaneously at approximately 12 months of age.

The MIT procedure was described by several authors in the 1990s (Mendoza et al., 1994; Kriens, 1997; Gunther et al., 1998). The aim of this method is minimal scarring. In this technique, incision lines are made along the borders of the cleft and along the medial side of the maxillary tuberosity. In CPS, incision lines are extended approximately one centimetre in the midline of the hard palate. Incisions are made according to the markings, and the mucoperiosteum is elevated from the nasal and oral sides of the palatal shelves in the hard palate. The hamulus is identified, through the lateral incisions, and the tendon of the tensor muscle is divided. The muscle aponeurosis and the nasal mucosa are released from the posterior border of the hard palate. Thereafter, the nasal layer, the uvula and the oral mucosa are sutured separately. Occasionally, in wide clefts, incisions have to be extended medial to the alveolar ridge to be able to suture the oral layer in the junction between the hard and soft palate (Figure 7).

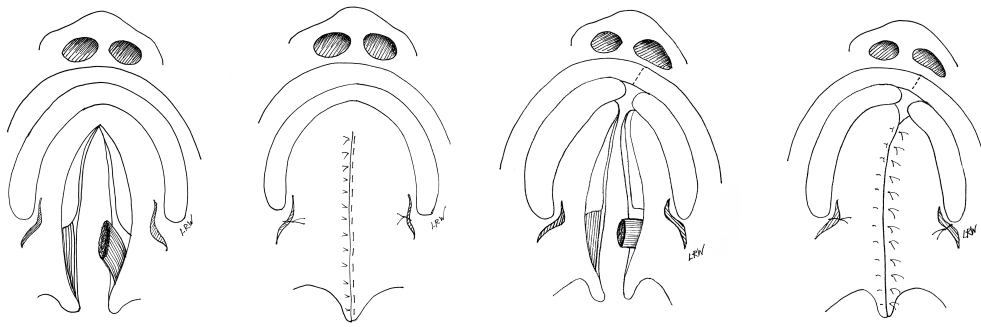


Figure 7. Cleft palate repair according to the MIT protocol in CPO to the left and in UCLP to the right. Illustrations by Liisi Raud Westberg.

The MITmr procedure was first introduced by Sommerlad (2003, 2006). In this method the surgeons use a microscope or magnification loupes. Incision lines are marked at the borders of the cleft and along the medial side of the maxillary tuberosity. In CPS, incision lines are extended approximately one centimetre in the midline of the hard palate. Incisions are made according to the markings and the mucoperiosteum in the hard palate is elevated. Dissection is continued posteriorly between the salivary glands and the muscle layer in the soft palate. First, the nasal layer and the uvula are sutured. The tensor aponeurosis is divided posterior to the hard palate and the muscles are dissected from the nasal mucosa and transposed posteriorly approximately 15 millimetres until the levator muscle is identified. The muscle bulges are then sutured in the midline. Finally, the oral layer is sutured and occasionally, in wide clefts, incisions have to be extended medial to the alveolar ridge to be able to suture the oral layer in the junction between the hard and soft palate (Figure 8).

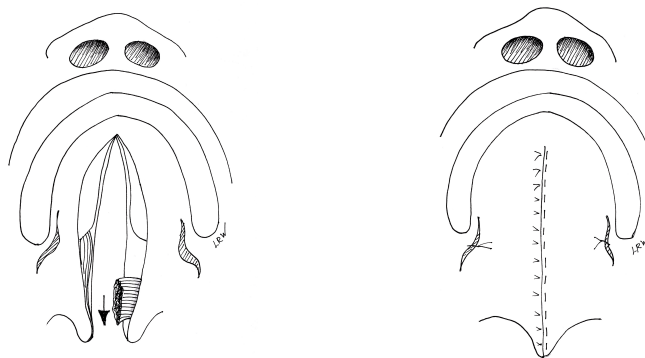


Figure 8. Cleft palate repair according to the MITmr protocol in CPO. Illustrations by Liisi Raud Westberg.

3.3 SPEECH SAMPLES AND AUDIO RECORDINGS

3.3.1 Audio recordings

The children were audio recorded at both 5 and 10 years of age, although the number of assessed recordings differed between the studies (Table 2).

Table 2. The number of audio recordings assessed in study I, II, III and IV at 5 and/or 10 years of age distributed by cleft type, reference group, the total number assessed at both 5 and 10 years and the total number.

Cleft group		Age		
		5 years	10 years	5 and 10 years
Study I	CPH	53		
	CPS	33		
	Reference	18		
Study II	UCLP	58	56	46
Study III*	CPH	36	57	31
	CPS	22	21	17
Study IV**	UCLP		10	
	Total	220	144	94


* Twenty-six of the audio recordings from study I were reassessed in Study III.

** Ten audio recordings from study II were used.

In study I and II were the children documented with either a Revox B77 M11 stereo tape recorder or a Sony DAT recorder (TCD-D8; Sony Corp., Japan) with a microphone (AKG D190E). In study III the same Sony DAT recorder was used or a H4nex Handy recorder (ZOOM Corp.) with an external microphone (NT4; Rode). The speech samples were digitized if needed and edited in the Soundswell program (sound file editor in Microsoft Windows system, MPC32d11, version 4.0 in study I and version 4.5 in study II). The audio samples were coded for blinding and each sample was saved in wav. format in a randomised order on a CD (study I), or on USB memory sticks (studies II and III). In study IV, speech samples from study II were used and edited in the freeware Audacity. The interviews in study IV were audio recorded with a H4nex Handy recorder (ZOOM Corp.) with an external microphone (NT4; Rode).

3.3.2 Speech samples

The speech samples consisted of repeated standardised sentences. Since the speech recordings lasted over a period of 22 years (1992 – 2014), the speech material were developed and refined over the years, however structured in the same manner. There were six to eight sentences containing only oral high-pressure consonants. Two to four sentences included oral and nasal consonants and one sentence had only nasal consonants (Table 3). When these sentences did not match the requirements of quality for the assessment, single words from picture–naming or counting 1–20 were used. This was the case in approximately 6% of the speech samples. In the focus groups sentence repetition together with counting were used for the listening procedure.

Table 3. Sentences used for repetition. The first section shows sentences used until 2003. The  second section shows sentences from SVANTE (Swedish Articulation and Nasality Test) used from 2004.

Sentence	Phonetic transcription	Translation
Pippis apa	pɪpɪs ʌpɑ	Pippi's monkey
Pippi åker polisbil	pɪpɪ ɔ:kɛɪ	Pippi is riding a police car
Titta på TV	tɪtɑ pɔ: tɛ:vɛ	Watch TV
I dag är det tisdag	ɪdɑ ɛ dɛ ti:sdɑ	Today is Tuesday
Kicki kokar potatis	kɪkɪ ku:kɑɪ putɑ:tɪs	Kicki is boiling potatoes
En kopp kaffe	ɛŋ kɔp kɑfɛ	A cup of coffee
Sissi sover	sɪsɪ sɔvɛɪ	Sisi is asleep
Solen lyser	sʊ:lɛn ly:sɛɪ	The sun is shining
Emil är inte snäll	ɛ:mɪl ɛ ɪntɛ snɛl	Emil is not nice
Mimmi å mamma e hemma	mɪmɪ ɔ mɑmɑ ɛ hɛmɑ	Mimmi and Mum are at home

Sentence	Phonetic transcription	Translation
Pippis apa piper	pɪpɪs ʌpɑ pi:pɛɪ	Pippi's monkey is squeaking
Bibbi bara jobbar	bɪbɪ bɑ:ɾɑ jɔbɑɪ	Bibbi is working all the time
Titti tittar på TV	tɪtɪ tɪtɑɪ pɔ: tɛ:vɛ	Titti is watching the TV
David och du leder	dɑvɪd ɔ dʉ lɛdɛɪ	David and you lead
Kicki kokar ägg	kɪkɪ ku:kɑɪ ɛg	Kicki is boiling eggs
Giggi vill väga guld	ɡɪɡɪ vɪl vɛɡɑ ɡʉld	Giggi would like to weigh gold
Sissi och Lasse sover	sɪsɪ ɔ lɑsɛ sɔvɛɪ	Sisi and Lasse is asleep
Fiffi får kaffe	fɪfɪ fɔɾ kɑfɛ	Fiffi will have coffee
Vivvi vevar	vɪvɪ vɛvɑɪ	Vivvi is cranking
Lollo lurar Ella	lɔlɔ lʉɾɑɪ ɛlɑ	Lollo is tricking on Ella
Svante vill inte ha vantar	sʉ ʌntɛ vɪl ɪntɛ hɑ vɑntɑɪ	Svante will not have gloves
Anki hämtar hinken	ɑŋkɪ hɛmtɑɪ hɪŋkɛn	Anki getting the bucket
Mimmi å mamma e hemma	mɪmɪ ɔ mɑmɑ ɛ hɛmɑ	Mimmi and Mum are at home

3.4 SPEECH ASSESSMENT

In study I, two external and two local SLPs from two cleft centres participated in the evaluation. In study II and III, cleft speech variables were independently rated from the recordings by three experienced external SLPs from three Swedish cleft treatment centres, all of whom specialised in cleft palate speech. The SLPs wore headphones and could listen to the speech samples an unlimited number of times. Once they had completed the evaluation, they could not go back and listen to the recording again. The listeners were blinded to the identity of the speaker, the surgical protocol and cleft type. For study IV, perceptual assessment for 10 speech samples in study II had been completed by three SLPs and the evaluation served as material for peers' descriptions of speech.

3.4.1 Speech variables and rating scales

The assessed speech variables related to cleft palate speech are presented in Table 4. The speech variables were rated at both 5 and 10 years of age on 5-point ordinal scales. Listeners could comment on any other type of articulation error detected. Regarding the rating of /s/-distortions, the listeners were also asked to describe the type of distortions as lateral, palatal or interdental articulation. In study I, hyponasality and nasal fricatives were also assessed.

Table 4. Speech variables and rating scales in study I, II and III.

	Hypernasality Hyponasality Weak Pressure Consonants	Audible nasal air leakage** Compensatory Articulation*** (Retracted oral articulation Glottal articulation, Nasal/pharyngeal fricatives) s-distortions****
	Degree of deviation	Frequency of occurrence
0	Normal speech/No deviation	No occurrence
1	Slight deviation	Single/occasional occurrences
2	Mild deviation	Some occurrences/occurs consistently on one sound/intermittently on several sounds
3	Moderate deviation	Frequently occurring
4	Severe deviation	Occurs always or almost always

* The scale values 0 and 1 were pooled to "green", 2 is presented as "yellow", and 3 and 4 to "red" in presentation of the results.

** Including audible nasal emission and velopharyngeal friction sounds.

*** Other types of articulation errors could be commented on as well.

**** Type of distortion was given, i.e. lateral, palatal or interdental.

In study I an estimation of velopharyngeal incompetence (VPI) was calculated based on the mean rank sum of the variables hypernasality, nasal air leakage and weak pressure consonants. An overall assessment of perceived velopharyngeal function and general impression of speech was rated using a 4-point scale in study II and III (Table 5).

Table 5. The 4-point scale for perceived velopharyngeal function and general impression of speech assessed in study II and III.

Scale Value*	Perceived velopharyngeal function and General impression of speech
0	Normal
1	Mildly incompetent/deviant
2	Moderate incompetent/deviant
3	Severely incompetent/deviant

* The scale values 0 and 1 were pooled to "green" in presentation of the results.

Finally, in study II and III, the SLPs were asked to decide whether, in their opinion, the child was in need of speech therapy and/or other additional treatment, such as velopharyngeal surgery.

3.5 FOCUS GROUPS INTERVIEWS

Focus groups interviews formed the basis for the data collection in study IV. In order to enable a thorough collection of the data, the interviews were performed by a moderator team with two SLPs – one handling the discussions and the other the recording equipment and taking notes. The information provided to the children about the speech examples was sparse. The moderator presented some examples of sentences in order to familiarise the children with the speech samples. A semistructured interview guide with open ended questions was used. The focus groups started with small talk about the voice and different dialects to introduce the topic and the subject cleft lip and palate was not mentioned. The peer comments were occasionally followed up by questions such as, "What did you hear? Did you understand?"

3.6 RELIABILITY

Fifteen percent of the audio recordings were duplicated to allow for repeated analysis and calculation of intra-rater reliability in study I. The corresponding figure in study II and III was 30%. Interrater-reliability was calculated from the assessments of all samples of all variables by all listeners. Intra- and inter-rater reliability was calculated with Spearman's rank correlation and Cronbach's alfa in study I, and as exact percentage agreement point-by-point for each variable and each listener in study II and III. The intra- and the inter-rater agreement was not considered acceptable on the scales, consequently one scale value difference was accepted as agreement (Table 6).

Table 6. Intra- and inter-rater reliability of the most prevalent variables in studies I, II, and III at 5 and 10 years of age.

	5 year of age						10 years of age			
	Study I		Study II		Study III		Study II		Study III	
	Spear- man's (Rho)	Cronbach's alfa	Mean % agreement within one scale value				Mean % agreement within one scale value			
	Intra	Inter	Intra (mean)	Inter	Intra (mean)	Inter	Intra (mean)	Inter	Intra (mean)	Inter
Hypernasality	.74	.84	87	78	100	75	97	92	96	93
Nasal air leakage	.75	.90	95	81	98	88	97	91	99	97
Retracted oral articulation	.79	.84	98	93	96	92	97	95	100	96
Glottal articulation	.86	.97	98	92	100	98	97	93	100	99
Velopharyngeal function	**	**	98	100	98	94	97	100	100	99
General impression	**	**	98	99	94	94	100	98	100	98

** The variable was not assessed in study I.

3.7 ANALYSIS

3.7.1 Statistical analysis

Non-parametric statistical analyses were chosen due the skewed distribution of data. The tests were carried out with IBM SPSS Statistics (version 12, 20, 23). Descriptive analyses were performed and prevalences presented in bar graphs. Differences within group between ages were tested with the Wilcoxon signed rank test (study II, III). Differences between two independent groups, i.e. surgical procedure and speech outcomes, between surgeons regarding the speech variables, and between the study group and the excluded group were tested with Mann-Whitney U-test (study I, II, III). The Kruskal-Wallis test was used to test for differences between the three groups (study I). Correlations between outcomes and independent variables were analysed with Spearman's rank correlation test (study I). The Chi-square exact test and Fisher's exact test were used for comparison of oronasal fistulas, hypernasality, and the number of

velopharyngeal flaps between the two surgical procedures (study I). A probability less than 5% was considered significant.

3.7.2 Qualitative content analysis

Qualitative content analysis was used in study IV in order to structure, condense and interpret the interview data. The interviews were transcribed verbatim by the first author. The analysis involved immersion in the data with repeated readings of the transcripts in order to identify and choose the units of analysis, including open coding, creating categories and abstraction. The categories and themes were continuously compared by the authors in order to ensure trustworthiness.

3.8 ETHICAL APPROVALS

Ethical approval was obtained from the Stockholm Regional Ethical Board: Study I: Dnr 188/03, study II and III: Dnr 2011/2065-31/3, study IV: Dnr 2014/18-31/3. All parents and children in study IV gave written informed consent to participate in the focus groups interviews.

4 RESULTS

In general, children with a cleft lip and palate demonstrated a progression in speech from 5 to 10 years of age, both concerning nasality and articulation. The graphs represent the percentage of children with different cleft types, judged to have moderately to severe deviation/incompetence or frequently or always/almost always occurring (scale value 3-4) speech errors. This level of difficulty is likely to require further examination and treatment of the VPF and of articulation errors. Four surgeons performed the operations in study I and two senior surgeons in study II and III respectively. No statistical difference in speech outcome was associated with the different surgeons.

4.1 ASPECTS OF FUNCTION

4.1.1 Nasality and perceived velopharyngeal function

Overall, the children with different cleft types presented with a high degree of hypernasality and audible nasal air leakage at 5 years of age, especially in the group operated on according to the MIT protocol. A gradual decrease with age was found, except for audible nasal air leakage in children with CPO. Velopharyngeal function was considered competent in most children at 10 years of age, but after the MIT protocol just over 40% of the children had received a velopharyngeal flap (Figure 9 and Figure 10).

Study I

Speech outcomes after the two different surgical techniques MIT and MITmr were compared at 5 years of age in children with CPO, and divided into CPH and CPS. The results showed no significant differences between the surgical methods for any of the evaluated speech variables, but significantly more children operated on with MIT had received a velopharyngeal flap. The cleft extent had an impact on speech, where children with CPH had significantly poorer speech regarding the variables of weak pressure consonants and glottal articulation than the children with CPS.

Study II

The speech outcomes in children born with UCLP treated with MIT were evaluated at 5 and 10 years of age. At 5 years, 19% of the children were perceived as having moderate to severe hypernasality and 17% had audible nasal air leakage, despite the fact that 12 children (17%) had received a velopharyngeal flap before the evaluation at 5 years. At 10 years of age, only 4% had moderate to severe hypernasality and 9% audible nasal air leakage. Another 18 children (26%) had received a velopharyngeal flap. In total, 30 children (43%) had a velopharyngeal flap before 10 years of age. The velopharyngeal function was perceived to be moderately to severely incompetent in 13% of the children at 5 years and in 7% at 10 years of age.

Study III

The speech outcomes in children born with CPO treated with MITmr were compared at 5 and 10 years of age. Twenty percent of the children showed moderate to severe hypernasality at 5 years of age, 11% in the CPH group and in the CPS group. At 10 years, the figure was 14%, 5% in the CPH group and 9% in the CPS group. Audible nasal air leakage was present in 40% of the children at 5 years and in 66% at 10 years of age and was more common in the CPS group than in the CPH group at both ages. This difference was statistically significant. In total 14% (12 children) received a velopharyngeal flap before 10 years of age, 10 children with CPH and two with CPS. At 5 years of age 20% of the children were perceived as having incompetent velopharyngeal function, 11% with CPH and 14% with CPS. At 10 years, the corresponding figure was 18%, 8% with CPH and 10% with CPS.

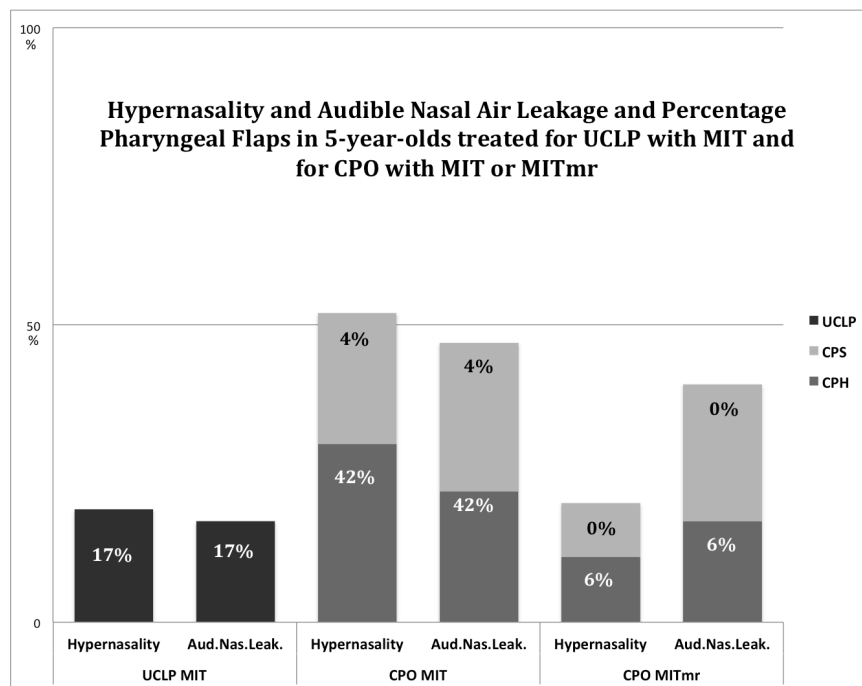


Figure 9. Hypernasality and audible nasal air leakage in 5-year-olds with UCLP or CPO treated with MIT or MITmr. The percentage of velopharyngeal flaps is presented in the bars.

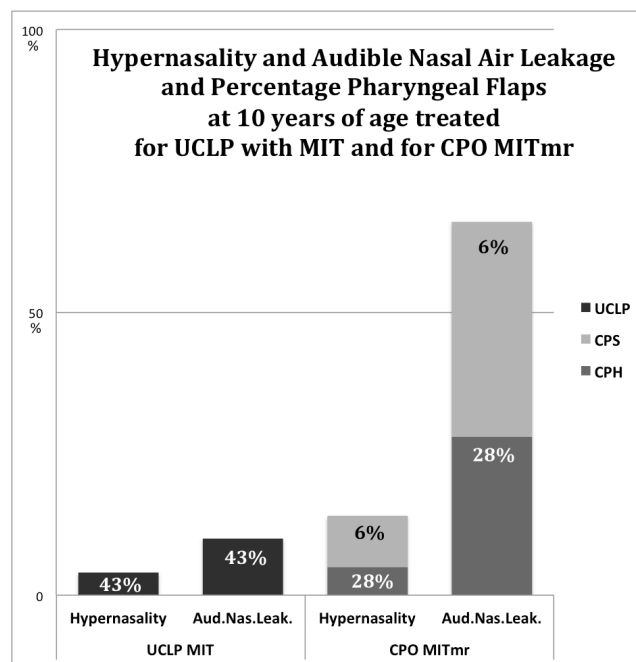


Figure 10. Hypernasality and audible nasal air leakage in 10-year-olds with UCLP or CPO treated with MIT or MITmr. The percentage of velopharyngeal flaps is presented in the bars.

4.1.2 Articulation

The main focus regarding articulation concerned compensatory articulation, such as retracted oral articulation and glottal articulation. The results showed that the CPO group had fewer articulatory difficulties than the UCLP group. At 5 years of age the most common articulatory error was glottal articulation in both the UCLP group and CPO group (study II and III). Furthermore, retracted oral articulation was present, above all in the UCLP group. The prevalence of a distorted /s/ was frequent in both the UCLP group and CPO group at 5 years. A general improvement in articulatory competence with age was noted for all cleft types (Figure 11).

Study I

Articulation deviances were mostly present in terms of retracted oral articulation (12%) and glottal articulation (20%). There was a significant difference between the CPH and CPS group regarding glottal articulation, which was more frequent in the CPH group than in the CPS group.

Study II

In the UCLP group treated with the surgical protocol MIT, retracted oral articulation occurred frequently or almost always (scale value 3–4) in 18% of the children at 5 years, while some occurrences (scale value 2) were found in 5% at 10 years. Glottal articulation was found frequently or almost always (scale value 3–4) in 12% of the children at 5 years, but decreased to 2% (one child) at 10 years. Distorted /s/ was common, but improved at 10 years of age.

Study III

In the CPO group treated with MITmr, the children displayed a low occurrence of compensatory articulation difficulties at both 5 and 10 years of age. Distorted /s/ was found, but an improvement was noted at 10 years of age.

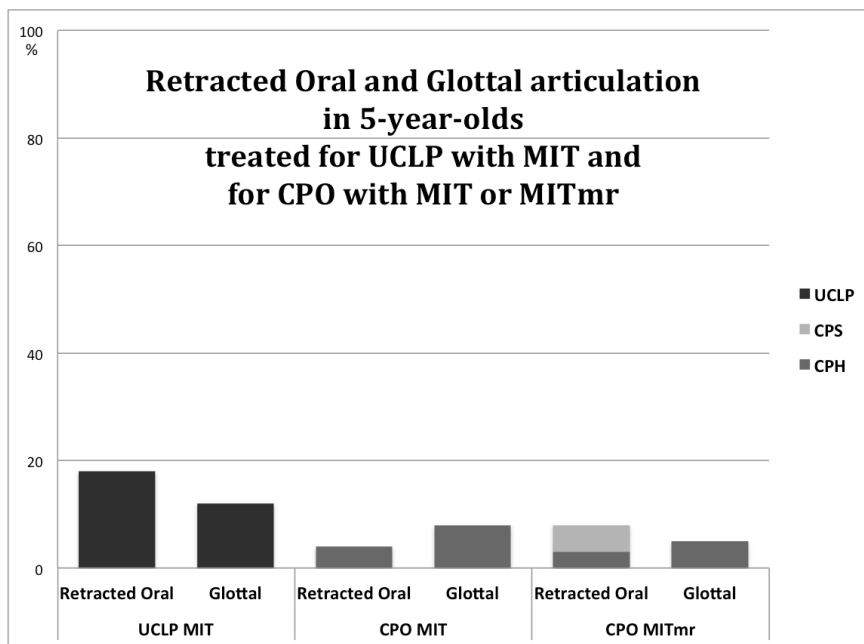


Figure 11. Retracted oral and glottal articulation in 5-year-olds with UCLP or CPO treated with MIT or MITmr.

4.1.3 General impression

An overall assessment of the general impression of speech was made in study II and study III. This variable could include both cleft-related speech deviances and comments on, for example, voice quality and distorted /s/-sounds. In both the UCLP group and CPO group the overall impression of speech improved with age. Ninety percent gave a normal impression of speech at 10 years of age in the UCLP group and between 90 to 95% in the CPO group.

4.2 ASPECTS OF ENVIRONMENT

The peers in the focus groups agreed with the SLPs regarding severe speech errors and also talked about possible consequences for the affected individual, both in terms of teasing/bullying, social acceptance and coping strategies. Peers easily detected and reacted to articulatory difficulties and speculated on the reasons why and how these errors could influence communication.

4.2.1 Nasality

Study IV

Ten-year-old peers were asked to describe speech and the possible communicative consequences for children with UCLP in their own words. The results showed that peers perceived more pronounced signs of VPI and referred to them in wording relevant to them. Describing hypernasality, they used words like "having a cold and a blocked nose". When hypernasality was accompanied by audible nasal air leakage, the reaction was stronger and the peers expressed thoughts about possible reasons for sounding strange and also had thoughts about consequences for the affected child, such as teasing, bullying and the possibility of being misunderstood. The peers questioned the accuracy of the speech samples and suggested that children with moderate hypernasality and severe audible nasal air leakage were kidding: "Are they pretending?" Speech samples with mild hypernasality and some occurrences of audible nasal air leakage were referred to as normal: "Sounds just like us".

4.2.2 Articulation

Study IV

Peers reacted strongly to articulation errors. The participants identified the same articulation impairments that were already described by the SLPs. Even minor articulatory difficulties were detected and commented on, especially retracted oral articulation, i.e. when a dental consonant was produced in a velar position, for example /t/ > [k]. They raised questions about the tongue and possible diseases causing the error. The peers stated that this sounded very childish, since you are not supposed to speak like this when you are 10 years old.

The children were sensitive to /s/-distortions, such as lateral or interdental /s/-articulation. They imitated and commented on the speech samples vividly and presented different solutions: "If you only could tighten the teeth a little".

5 DISCUSSION

In this project, the speech outcomes in children born with UCLP and CPO were investigated at 5 and 10 years of age. All children were treated according to one of two surgical protocols, MIT and MITmr, in a one-stage procedure at approximately 12 months of age. In addition to the evaluation of aspects of function, environmental aspects were included. Ten-year-olds were invited to describe speech in children born with CLP and their thoughts about possible consequences regarding sounding different were explored.

5.1 RESULTS

The main findings were related to nasality, articulation and secondary surgery, as well as to descriptions and comments from peers, participating in focus groups. Speech improved with age, although some deviances remained at 10 years. The speech impairments found were signs of velopharyngeal insufficiency and articulatory difficulties. Focus group interviews with peers verified that they perceived the speech characteristics assessed by experts, and in addition, had thoughts on consequences, emotional reactions and associations.

A high occurrence of hypernasality and audible nasal air leakage was found at 5 years of age, especially in the group operated with MIT. In the CPO group in study I, 50% of the children had mild to severe hypernasality with no significant difference between the group MIT or MITmr. However, the group treated with MIT had a significantly higher frequency of velopharyngeal flaps. Children with a more extensive cleft (CPH) had a higher prevalence of hypernasality (30%) compared to those with less extensive (CPS). A comparable high occurrence of hypernasality was also found in the CPS group (20%). The result in the CPH group was similar to what was found by Persson et al. (2002), who evaluated speech after a two-stage protocol. However, 6% in the CPS group were rated as having moderate hypernasality. In fact, there was no difference between the CPS group and the reference group in the study by Persson et al. (2002). In the other group treated with MIT in the present project – that is, children with UCLP – the results showed that 19% of the children were perceived as having moderate to severe hypernasality at 5 years of age, in spite of the fact that

17% of the children already had received a velopharyngeal flap. Few studies report high numbers of velopharyngeal flap operations before age 5 years. However, one centre in the Scandcleft project reported 23% (Lohmander et al., 2016). Additional treatment with a velopharyngeal flap was presumably the reason for the improvement of hypernasality at 10 years of age in study II in the present project. Thus, the velopharyngeal function was considered competent in most children at 10 years of age, but after the MIT protocol, over 43% of the children with UCLP had received a velopharyngeal flap. In comparison, Sell (2005) reported speech outcomes from 238 children born with UCLP at age 5, and from 218 children at 12 years of age treated with various surgical protocols, 18% had consistent mild to severe hypernasality at both ages. By adding the patients who had undergone velopharyngeal surgery with to those who presented consistent hypernasality, the authors concluded that 29% of the 5-year-olds and 32% of the 12-year-olds had an inadequate primary repair in terms of velopharyngeal function. The corresponding figures in the present study were higher with 36% at age 5 years and 47% at 10 years of age. As in the present project, a gradual improvement in speech was reported by Lohmander et al. (2012), evaluating a two-stage procedure in UCLP-patients. At 5 years of age 25% had more than mild hypernasality and at age 10 this figure was reduced to 6%. The estimated primary VPI was low, i.e. around 15% including the number of velopharyngeal flaps.

The peer groups did not comment on mild hypernasality or some occurrences of nasal air leakage, but paid attention to and described moderate to severe hypernasality with terms for nasality like having a blocked nose: "Kind of stuffy nose" and "I think he sounded snotty, as having a cold". These statements are in agreement with untrained adult listeners' descriptions of nasality (Brunnegård et al., 2009.) When the hypernasality was accompanied with nasal air leakage the peers interpreted that as: "It sounds like he is blowing his nose" and "It is like he is holding his nose and talking at the same time". The fact that the peers did not reflect on mild/some occurrence in these variables confirmed the validity of the decision to pool the scale steps (0–1) in the assessments by the experts.

Based on the findings in study I and II, the interest in speech outcomes after treatment with MITmr was the focus in the next study on speech outcomes in CPO (study III). Although a low number of velopharyngeal flap surgeries were presented in the group and a comparable low occurrence of hypernasality was found at both 5 and 10 years of age, the occurrence of audible nasal air leakage was high, already at age 5 years (40%) and increased to 64% at age 10. The high occurrence was found in both cleft types. It is reasonable to believe that the primary palatal surgery was not effective enough to accomplish a satisfactory velopharyngeal

function. However, a decrease of the adenoid and growth of the nasopharynx might also contribute to the increased audible nasal air leakage (Finkelstein et al., 1996; Sell & Pereira, 2011). A higher number of secondary velopharyngeal surgery would be needed in order to normalise the speech.

The number of velopharyngeal flap operations was higher in the UCLP group compared to the CPO group. This was clear for the groups operated on with the MIT protocol. Whether or not this holds true also after treatment with the MITmr protocol can not be determined from the current project since the MITmr was not evaluated in the UCLP group. Based on the results in the CPO group, the results also indicate the need for a higher number of secondary velopharyngeal surgeries after MITmr, which brought a change in surgical protocol at the Stockholm centre. Interestingly, there are studies where secondary velopharyngeal surgery had not been performed at all in children with a non-syndromic CPO (Persson et al., 2002). Timmons et al. (2001), reported no velopharyngeal flaps in the UCLP group, whereas 37% had velopharyngeal flaps in the CPO group. In studies of patients with UCLP, which reported long-term results around 20% of the children had received a velopharyngeal flap (Karling et al., 1993; Farzaneh et al., 2008; Havstam et al., 2008). This is comparable to the figures at 5 and 10 years of age in the study by Sell et al. (2001), but significantly lower than in the present project.

Compensatory articulation was found in both the UCLP and the CPO group. However, the CPO group had less articulatory difficulties than the UCLP group and children with CPS exhibited only minor deviances. This is in agreement with the results reported by Timmons et al. (2001) and Persson et al. (2006). Interestingly, both retracted oral and glottal articulation were present at 5 years of age in study I and II, with slightly higher occurrence of glottal articulation (20%) compared to retracted oral articulation (12%) in CPO and the reverse in UCLP. A high occurrence of retracted oral articulation after a one-stage protocol is not commonly reported, whereas this has been considered the main speech problem in the young children treated with a two-stage procedure (e.g. Lohmander et al., 2012). It could be that this type of articulation error, often called "backing", has not been considered cleft-related and has therefore not been reported.

Even though the speech is commonly reported as improved at age 10 years, there might be persistent speech difficulties even in 10-year-olds. For example, in two studies on 10-year-olds born with a unilateral cleft lip and palate, between 34% and 46% of the children had impaired speech (Brunnegård & Lohmander, 2007; Hortis-Dzierzbicka et al., 2012). Furthermore, in the study on 12-year-olds born with a unilateral cleft lip and palate, 19% had

speech that was different enough to provoke comment according to the assessments made by the SLPs (Sell et al., 2001). In the present studies, a rather low occurrence of articulation errors were found together with distorted /s/-articulation, which was enough to raise comments in the peer groups. They were sensitive to impaired articulation and commented on both retracted oral articulation and /s/-distortion. These difficulties were associated with having anatomical differences and could, according to peers result in being perceived as childish and at risk for bullying and having a hard time in the classroom. Retracted oral articulation (/t/ articulated as [k]) was described as, for example, "It sounds as if he has folded up his tongue when he speaks", and, "You can hear that he has difficulties with letters, where you use your tongue", and, "He can't speak". Peers also easily detected slight or mild distorted /s/-articulation, with lateral and interdental production: "He is lisping a lot" and concluded, "If you are going to present [your work in the class room], it can be really hard". These comments show that deviant speech in isolation is enough to provoke negative comments and reflections on the person being different. Earlier research has reported teasing and bullying in children with cleft. Hunt et al. (2006) found that 62% of the participants with CLP reported having been teased, compared with 22% in the control group. The majority of teasing/bullying was related to the subject's facial appearance (55%) or speech (34%). Since many of the previous studies have focused on appearance (e.g. Marcusson et al., 2002; Hunt et al., 2006;), the impact of speech was important to investigate. The findings of this study showed that children with deviant speech risk negative comments from peers and point to a need for intervention programs to counteract bullying and social rejection at schools.

The general impression of speech evaluated in study II and III improved with age, indicating a positive progression in the speech variables assessed. A possible reason for not being rated as giving a normal impression at 10 years of age might be the /s/-distorsions. These articulation errors are also prevalent in typically developed children, and it is assumed that if they had been included as well, the perceived impression would probably have been affected in the same way. When the participants in study IV commented on speech in 10-year-olds assessed as having normal speech, they sometimes referred to themselves: "It sounds a bit cocky to say that she talks like we do", and, "It sounded completely normal".

5.2 METHODOLOGICAL CONSIDERATIONS

The risk of missing data is due to the lack of examinations and audio recordings that is common in longitudinal studies (Karnell & Van Demark, 1986; Hardin-Jones et al., 1993; Lohmander et al., 2012). This was also the case with both study II and study III, but the amount of audio recordings at both ages was reasonably high and the evaluation at both ages as well as a comparison between ages could be performed. The children in the studies were born in 1987 and 2004. During this time, both technical equipment and clinical routines for documentation had developed. However, the assessed variables were the same. The decision to include or exclude children with diagnosed syndromes or additional malformations in studies of speech outcomes has been recognized and discussed in previous studies (Persson et al., 2002; 2006). In the present studies, these children were excluded because of the possible interference of confounding factors, such as neurodevelopmental delay. The reference group in the present study (study I) performed in accordance with the Swedish normative data at 5 years of age, both regarding nasality and articulation (Lohmander et al., 2016).

Two different surgical protocols, MIT and MITmr, were evaluated in terms of speech outcomes. The MIT method was performed until 1997, while the protocol according to MITmr was introduced at the end of 1997. Consequently there was a transition period, when both of the techniques were in practice. The children treated during this period were not included in order to avoid the impact of a learning curve. To ensure that the children were correctly analysed regarding surgical protocols, all surgical records with detailed descriptions of the techniques used were analysed by a plastic surgeon on the team. There were no differences regarding speech outcomes between the three surgeons, indicating consensus on surgical protocols.

Sentence repetition was used as speech samples in this project. Klintö et al. (2011) reported that sentence repetition is a speech material reflecting coherent speech with high reliability and validity. An advantage is that the speech material is standardised and the assessors know the target sound. According to Sell (2005), blinded independent analysis of speech data is recommended as the gold standard when reporting research outcomes. In study I, four listeners blinded to surgical protocol and cleft type participated, and in study II and III three listeners respectively blinded to identity and/or cleft type evaluated the speech samples. The reliability was secured by repeated assessment within and between listeners. In study IV, sentence repetition and counting from study III was used as speech samples.

Low agreement was most prominent in the variables of hypernasality and nasal air leakage at 5 years of age. This has been reported previously (e.g. Karling et al., 1993; Timmons et al., 2001; Brunnegård & Lohmander, 2007; Lohmander & Persson 2008) and constitutes a problem in the evaluation of cleft palate speech. On the other hand, the disagreement was primarily for the lower scale values, which are mostly left without any further treatment or examination. In addition and interestingly, this was in accordance with the descriptions from peers who paid no attention to minor symptoms of VPI, but observed and commented on more prominent hypernasality. Havstam et al. (2011) concluded that the velopharyngeal function appears to have a limited impact on the child's communication attitude, at least when signs of impairment are not pronounced.

When evaluating data from a long period of time, limitations in both background information and analysis were found. This project reported on speech outcomes at 5 and 10 years of age (study II, III) and during this time routines for documentation changed, as well as the technical equipment and storage of recordings. Thus, unfortunately, not all children were assessed at both ages. The clinical routines regarding hearing and otological data were not well established for the time of the included studies. Consequently, not all children had a hearing test at 5 and 10 years. A main weakness was also the lack of data concerning speech therapy, both about the amount given and about description of the content and methods provided.

The international Ethical Research Involving Children project, ERIC, initiated by UNICEF (Graham et al., 2013) seeks to ensure the dignity and human rights of children in research. Emphasis is placed on the value, importance and legitimacy of research that captures the views and perspectives of children and young people. Youth focus groups have been engaged in formulating questions in different fields of research (Peterson-Sweeney, 2005), and is considered an explorative method for discovering how young people think about various issues in their environment (Krueger & Casey, 2009). In study IV, the participating peers gave written consent to participate in a study "listening to and describing voices". Focus group interviews were the basis of this study and qualitative content analysis was used as the method for evaluation, which was recommended for interpreting data from focus groups (Krippendorff, 2013; Elo & Kyngäs, 2008; Graneheim & Lundman, 2004), especially when the study deals with an unknown topic. The analysis resulted in three interlinked categories, which encompassed different aspects of speech, personality, and social implications: descriptions of speech, thoughts on causes and consequences, and emotional reactions and associations covering feelings and reactions in the affected individual as well as in the

environment and among peers. The results showed that the subject engaged peers and highlighted the fact that traditional assessment of speech outcome needs to be complemented with aspects from real life. Treatment should be based on what is relevant to the afflicted child taking their social context into consideration.

6 CONCLUSIONS AND CLINICAL IMPLICATIONS

- Nasality and articulation improved with age, except for nasal air leakage in the CPO group, assuming that a higher number of velopharyngeal flaps might be required at a later age.
- Minor signs of nasality and/or VPI were not noted and commented on by peers. This indicates that the mild degree or single occurrences of hypernasality and audible nasal air leakage needs no further clinical intervention.
- Articulation difficulties were not a prominent problem in this cohorts. Nevertheless, peers were sensitive even to minor articulatory errors. Consequently, in order to provide effective treatment intervention, it is important to ask the affected child about their communicative experience and how they feel about speaking in various situations.
- The clinical routines for follow up and documentation of speech and audio recordings have been reorganised, and hearing tests and otological data are collected in a more structured manner.
- The MIT surgical protocol resulted in a high number of velopharyngeal flaps compared to the MITmr procedure, and secondary surgery was more prevalent in the UCLP group than in the CPO group. In the CPO group, children with CPH exhibited a higher number of velopharyngeal flaps than the CPS group, irrespective of surgical method.
- The MITmr protocol is a complicated surgical procedure, which demands an extensive muscle reconstruction, and therefore, takes long time to perform and initially to learn. The speech results after the MITmr surgical protocol were not convincing enough. Consequently, the surgical procedure has been changed by the Craniofacial Team in Stockholm, where a two-stage protocol is nowadays performed.

Children born with a cleft lip and palate are at potential risk for speech impairment. In order to implement the best possible care, a continuous evaluation of surgical protocols is necessary as well as to provide results from speech outcomes at different ages. Furthermore, a wider focus on outcomes should be adopted, and include aspects other than function.

7 FUTURE STUDIES

In agreement with the guidelines from for example, the International Consortium for Health Outcome Measurement, ICHOM, there is a need to broaden the perspective of outcome measures in cleft care (www.ichom.org/medical-conditions/cleft-lip-palate/). A focus on patient-centered results, including patient and parent-reported speech measures, needs to be included when evaluating treatment outcome.

Studies of speech intervention is required to identify the methods that are effective, including their impact on other aspects of children's communication and social participation and well-being.

Environmental aspects needs to be expanded, and based on the findings from the present project regarding peers perception and descriptions of speech, larger groups should be included. In order to study if age affects the way a person perceives and describes speech, children from different age groups could be included. An assessment tool adapted to young listeners could add more information about the possible impact of speech impairment in larger groups.

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