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**SPEECH AND EXPRESSIVE LANGUAGE  
IN SWEDISH-SPEAKING CHILDREN  
WITH UNILATERAL CLEFT LIP AND PALATE**

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# SPEECH AND EXPRESSIVE LANGUAGE IN SWEDISH-SPEAKING CHILDREN WITH UNILATERAL CLEFT LIP AND PALATE

## THESIS FOR DOCTORAL DEGREE (Ph.D.)

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## ABSTRACT

**Introduction and aims:** A cleft palate may hamper development of speech and expressive language. Expressive language, including phonology, has been sparsely explored in children with cleft lip and palate, and extended knowledge is essential in order to improve intervention for this patient group. The aims of the project were to assess the effectiveness of different speech materials used when evaluating cleft palate speech, to assess the development of speech and expressive language in children born with unilateral cleft lip and palate, and to identify variables in early speech production, which may be associated with later expressive language.

**Material and methods:** Thirty children with unilateral cleft lip and palate, treated with three different methods for primary palatal surgery, and 20 children without cleft lip and palate participated. Speech was longitudinally documented at 18 months, 3 years, and 5 years of age. The effectiveness of four different speech materials for assessing cleft palate speech was explored. Articulation/phonology at 3 and 5 years were studied, and the outcomes were correlated with earlier outcomes of consonant production. Speech and phonology in children treated with different methods for primary palatal surgery were assessed. Expressive language in narrative retelling was assessed and the outcomes were compared with outcomes of articulation/phonology.

**Results:** The best speech performance and reliability were achieved in single word naming. The reliability in sentence repetition was good, and speech performance was equally good as in conversational speech. The group with unilateral cleft lip and palate displayed deviant phonology at 3 and 5 years of age, compared with peers without cleft palate. Measures of consonant production at 18 months of age correlated significantly with the outcomes at 3 years of age, and there also was a significant correlation between the outcomes at 3 and 5 years of age. The results indicated a two-stage palatal surgery with hard palate closure as late as 3 years of age to be disadvantageous for the development of speech and phonology. At 5 years of age, a larger proportion of the children with unilateral cleft lip and palate than peers without cleft had problems retelling information and these problems were not related to surgical method, gender, or articulatory/phonological competence.

**Conclusions:** Word naming, in combination with sentence repetition, is recommended for evaluation of cleft palate speech when best performance and performance in coherent speech are assessed. Many children with unilateral cleft lip and palate have phonological problems at up to 5 years of age. It seems possible to identify children at risk for impaired phonology at earlier ages for possible prevention of persistent problems. Two-stage palatal surgery with hard palate closure as late as 3 years of age should be avoided since it may hamper phonological development. In addition, many children with unilateral cleft lip and palate have problems retelling information at 5 years of age, unrelated to articulatory and phonological ability, and may be in need of further language intervention.

## SAMMANFATTNING

**Introduktion och syfte:** En gomspalt kan hämma utvecklingen av tal och expressivt språk. Expressivt språk, inklusive fonologi, har endast blivit sparsamt utforskat hos barn med läpp-käk-gomspalt, och kunskaperna behöver utökas för att behandlingen ska kunna förbättras. Syftet med projektet var att undersöka hur tillförlitliga olika talmaterial är för utvärdering av tal hos barn med gomspalt, att undersöka utvecklingen av tal och expressivt språk hos barn födda med enkelsidig läpp-käk-gomspalt, och att identifiera variabler i tidig talproduktion som kan ha samband med senare expressivt språk.

**Material och metoder:** Trettio barn med enkelsidig läpp-käk-gomspalt, behandlade med tre olika metoder för primär gomslutning, och 20 barn utan läpp-käk-gomspalt deltog. Talet dokumenterades longitudinellt vid 18 månaders, 3 års och 5 års ålder. Fyra talmaterial för bedömning av tal vid gomspalt utvärderades. Artikulation/fonologi vid 3 och 5 års ålder studerades, och sambandet med tidigare resultat avseende konsonantproduktion undersöktes. Tal och fonologi hos barn behandlade med olika metoder för primär gomslutning jämfördes. Expressivt språk vid återberättande undersöktes, och samband med artikulatorisk/fonologisk förmåga studerades.

**Resultat:** De bästa talresultaten och den högsta reliabiliteten uppnåddes vid benämning av enstaka ord. Reliabiliteten vid meningsrepetition var god, och talresultaten var jämförbara med dem i spontantal. Barnen med enkelsidig läpp-käk-gomspalt hade som grupp avvikande fonologi vid 3 och 5 års ålder, jämfört med jämnåriga utan spalt. Mått på konsonantproduktionen vid 18 månaders ålder korrelerade signifikant med resultaten vid 3 års ålder, och det var även ett signifikant samband mellan resultaten vid 3 och 5 års ålder. Resultaten indikerade att det är negativt för utvecklingen av tal och fonologi med tvåstegslutning om hårda gommen sluts så sent som vid 3 års ålder. Vid 5 års ålder hade en större andel barn med enkelsidig läpp-käk-gomspalt än jämnåriga utan spalt problem att återge information, och svårigheterna hade inget samband med kirurgisk metod, kön eller artikulatorisk/fonologisk förmåga.

**Slutsatser:** Ordbenämning i kombination med meningsrepetition rekommenderas för utvärdering av tal hos barn med gomspalt, om man vill undersöka barnets bästa prestation och sammanhängande tal. Många barn med enkelsidig läpp-käk-gomspalt har fonologiska svårigheter upp till 5 års ålder, och barn i riskzonen för fonologisk språkstörning verkar kunna identifieras i tidigare ålder, för att om möjligt förebygga långvariga problem. Primär gomslutning i två steg med slutning av den hårda gommen vid 3 års ålder bör undvikas, eftersom den kan hämma den fonologiska utvecklingen. Dessutom har många barn med enkelsidig läpp-käk-gomspalt problem med att återge information vid 5 års ålder, som inte har samband med artikulation och fonologi, och kan vara i behov av ytterligare logopedinsatser.

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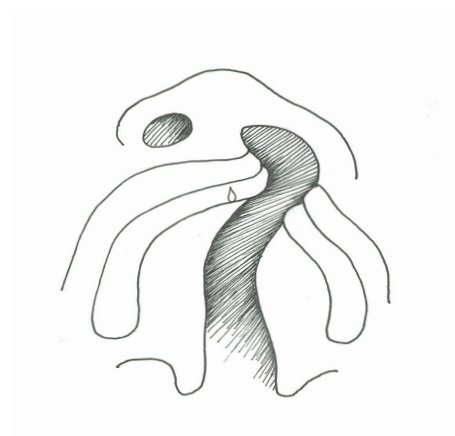
# 1 INTRODUCTION

## 1.1 CLEFT LIP AND PALATE

### 1.1.1 Incidence and implications

Cleft lip and palate is a congenital malformation that arises during the 5th to 12th week of pregnancy, when the facial parts are expected to grow together. If the fusion does not occur, a child is born with a cleft. The incidence in Sweden is about 2/1000, which results in 150–200 new-borns every year with some type of cleft lip and palate (Hagberg et al., 1998). About one third are born with unilateral cleft lip and palate, with a cleft either on the right or the left side of the lip and alveolus in combination with a cleft palate (Figure 1). Depending on the type and extent of the cleft, it may affect eating, ear function, hearing, babbling and speech, development of the teeth and jaw, and also facial appearance. In order to give a child prerequisites for optimal development of these structures and functions, the cleft is surgically closed.

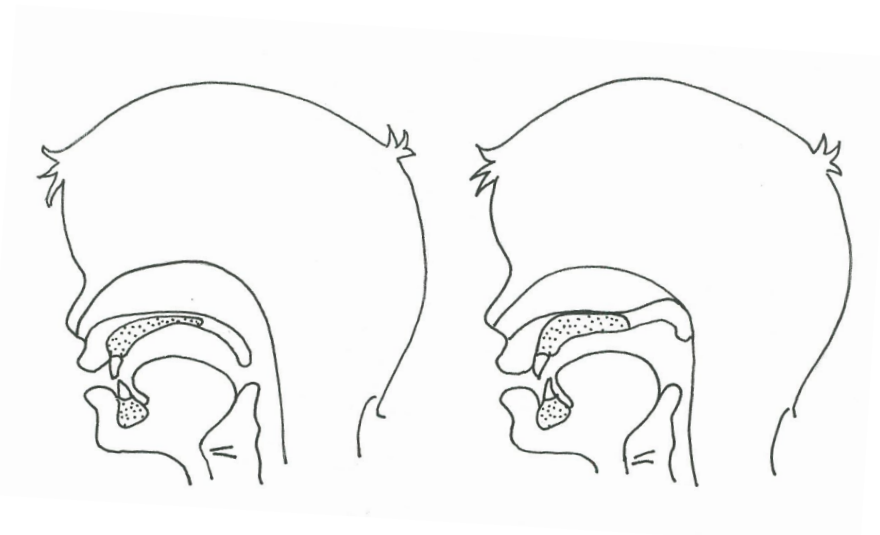
If there is a cleft in the palate, speech therapy and/or secondary speech improving surgery may be necessary. Orthodontic treatment is also often needed. In addition, children born with cleft palate are more affected by liquid trapped in the middle ear, i.e., otitis media with effusion, than are peers without cleft palate, which may result in hearing impairment. For optimal treatment results, plastic surgeons, orthodontists, speech-language pathologists, and audiologists interact in teams.



*Figure 1.* Unilateral cleft lip and palate seen from below. Illustration by Liisi Raud Westberg.

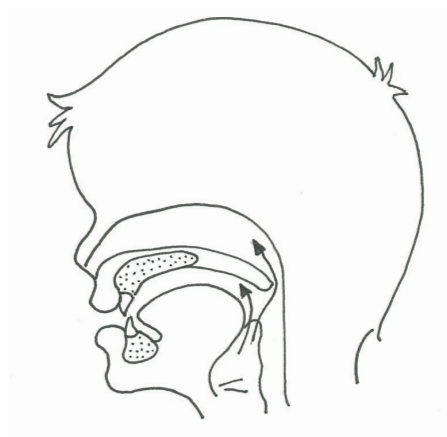
### 1.1.2 Primary palatal surgery

The anterior hard palate consists of bone. Posteriorly, the hard palate merges into the soft palate, which consists of muscles. A prerequisite for normal speech production is that the passage between the oral and nasal cavity can be separated, using the soft palate (velum) and pharyngeal walls (pharynx) (Figure 2).



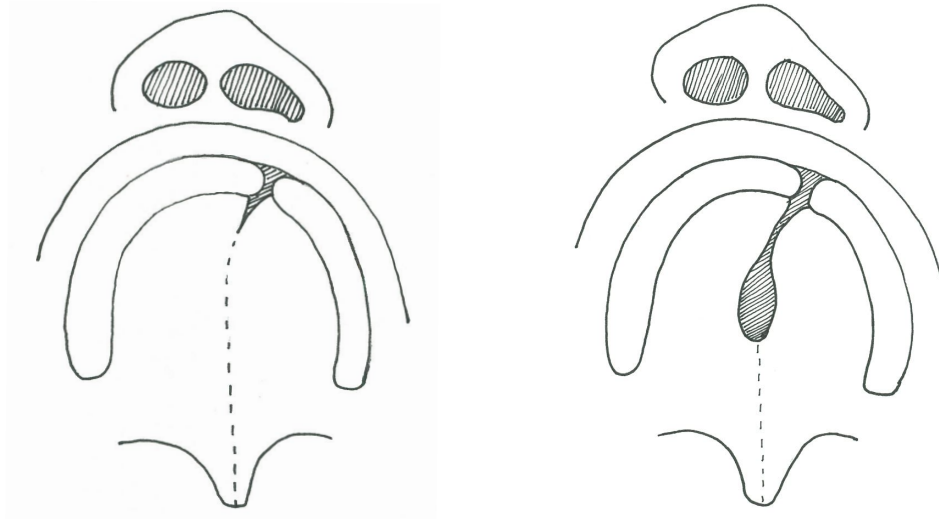
*Figure 2.* To the left, velopharynx during breathing. To the right, sufficient velopharyngeal closure. Illustration by Liisi Raud Westberg.

Even after a completed primary palatal surgery, the velopharyngeal closing mechanism may be insufficient due to a short palate, reduced mobility of the pharynx/velum, or disproportion between length of the palate and throat depth (Figure 3).



*Figure 3.* Insufficient velopharyngeal closing mechanism. Illustration by Liisi Raud Westberg.

Also, a postoperative fistula, due to surgical failure, or a deliberately un-operated cleft in the alveolar ridge or the hard palate, left to be closed at a later age, may result in oro-nasal coupling (Figure 4).



*Figure 4.* To the left, an un-operated cleft in the alveolar ridge. To the right, an un-operated cleft in the alveolar ridge and the hard palate. Illustration by Liisi Raud Westberg.

The main goal of primary palatal surgery is to obtain appropriate palatal length, velopharyngeal competence, absence of postoperative fistulas, and undisturbed mid-facial growth. According to a common opinion, speech development benefits from palatal closure as early as possible (Peterson-Falzone, 1996; Rohrich et al., 2000). On the other hand, facial growth may benefit from delayed palatal closure (Rohrich et al., 2000; Friede, 2007). Irrespective of the timing of the palatal operation, surgical techniques that result in a denuded maxillary bone may have a negative impact on facial growth (LaRossa, 2000).

In 1859, van Langenbeck developed a technique for palatal repair still used today, where the palate is closed with medial mucoperiosteal flaps without lengthening (Goldwyn, 1969). Today, the van Langenbeck technique is also used together with techniques providing a well-functioning muscle sling and increased palatal length (LaRossa, 2000). In the 1930s, pushback palatoplasty was developed for increased palatal length (Veau, 1931; Wallace, 1987). The disadvantage of pushback palatoplasty is that bone is exposed (LaRossa, 2000). In addition, the frequency of postoperative fistulas is high with this technique (Cohen et al.,

1991). In order to bypass these problems, two-flap palatoplasty (Bardach and Salyer, 1986) and double opposing z-palatoplasty (Furlow, 1986) were developed in the 1960s and 1970s. Thereafter, techniques for muscle repositioning and intravelar veloplasty have been developed in order to improve the velopharyngeal competence. Sommerlad (2003) suggested a technique for radical repositioning of the velar musculature and tensor tenotomy, resulting in decreased need for secondary speech improving velopharyngeal surgery.

To facilitate mid-facial growth, a two-stage palatoplasty was developed by Schweckendiek with early soft palate closure together with lip closure at 4 to 6 months of age and hard palate closure at 12 to 15 years of age (Schweckendiek and Doz, 1978). Others have modified both the technique and timing of surgery in the two-stage approach. For example, Rohrich et al. (1996) suggested a two-stage repair with early soft palate closure in combination with hard palate closure at 15 to 18 months of age. Early soft palate closure is believed to promote the development of speech sounds (Willadsen and Albrechtsen, 2006).

Swedish children born with unilateral cleft lip and palate are currently treated with a primary lip plasty with simultaneous correction of the nasal cartilages at 3 to 6 months of age. The palate is either closed in one stage at 12 to 15 months of age, or in two stages with soft palate closure in connection with lip plasty and hard palate closure at about 2 years of age. The delayed closure of the hard palate is based on the concept that facial growth will thereby be promoted. In the mixed dentition at 8 to 9 years of age, the residual cleft in the alveolar ridge is closed by a cancellous bone transplant from the iliac crest or tibia.

## **1.2 SPEECH AND EXPRESSIVE LANGUAGE**

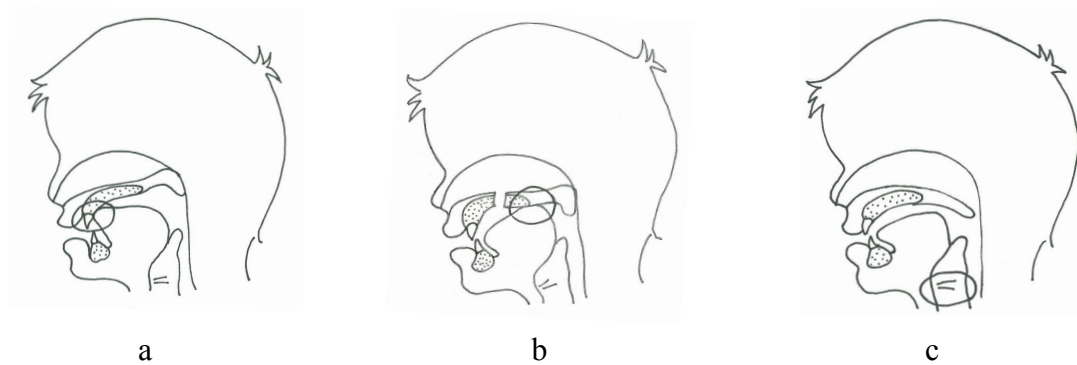
Speech can be defined as the verbal means of communicating language. Prerequisites for optimal speech are a well-functioning voice, articulation, resonance, and fluency. In children born with cleft palate, primarily articulation and resonance may be impaired. Language ability can be divided into four domains: phonology (i.e., the contrastive use of speech sounds and the phonotactic rules for combining phonemes, specific for different languages), grammar, semantics (i.e., content aspects of language and the meaning of words), and

pragmatics (i.e., overall communication skills and language use). Expressive language usually refers to the ability to express oneself in speech and writing. The focus in this thesis was speech, in terms of articulation and resonance, and aspects of expressive language in children born with unilateral cleft lip and palate.

### 1.2.1 Why a cleft palate may hamper speech production

If there is unwanted oro-nasal coupling, the nasal cavities will be involved in speech production. As a consequence, speech difficulties may arise, such as hypernasal resonance, audible nasal air leakage, and weak or nasal realized oral consonants. These cleft speech characteristics are regarded as passive (Harding and Grunwell, 1998).

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/) these consonants may be produced at a place behind the oro-nasal coupling (Henningsson and Isberg, 1990). This compensatory strategy is regarded as an active process (Harding and Grunwell, 1998). One example of this is retracted oral articulation. If there is an oro-nasal opening in the palate or the alveolar ridge due to a residual cleft or postoperative fistula, consonants normally produced anteriorly, such as in Swedish the dental stop /t/ (Figure 5a), may be produced at a place behind the oro-nasal opening, usually at a velar place, and, thus, be realized as, for example, the velar stop /k/ (Figure 5b) (Henningsson and Isberg, 1990). Another example of an active compensatory strategy is glottal articulation. If consonant articulation becomes weak due to inadequate function in the velopharynx, the consonants may be produced at the vocal cord level (glottis). Oral consonants are then realized as non-oral glottal consonants (Figure 5c). Active processes may persist even after a successful primary palatal repair (Harding and Grunwell, 1998).



*Figure 5. (a) Correct place of dental/alveolar articulation. (b) Articulation retracted to velar articulation place. (c) Articulation retracted to vocal cord level. Illustration by Liisi Raud Westberg.*

### 1.2.2 Articulation and phonology

The production of speech sounds in the vocal tract and in the oral and nasal cavities can be phonetically described in articulatory terms. For this purpose, phonetic transcription may be used in order to analyse the separate units of speech in a linear sequence. Although speech does not consist of separate units but rather of elements joined in a continuous flow interacting through co-articulation, phonetic transcription is useful in order to identify and prioritize which aspects of speech need to be focused on (Heselwood and Howard, 2009).

In speech pathology, phonetic or articulatory terms have been applied to articulatory deviances with origins in anatomical or motor deficits. In the late 1960s, the concept of phonology was introduced into speech pathology and speech therapy, and, in the 1980s, it was fully established (Grunwell, 1985). A phonological description of speech deals with the contrastive use of speech sounds and the phonotactic rules for combining phonemes. Phonological terms may also describe speech problems assumed to be related to a deficit on a cognitive-linguistic level (Nettelbladt, 1983; Dodd, 2005). The distinction between articulation and phonology is seen as important, although not always easily made, since articulatory processes may be incorporated in a child's phonology, i.e., influencing the organization of phonological patterns and the reception of new auditive information (Locke, 1993).

Currently, in speech language-pathology, classification of surface error patterns in speech production is commonly used to determine the level of deficit (Dodd, 2005; Netelbladt,

2007b). Phonological processes, i.e., processes resulting in a loss of phonemic distinction and a contrastive function of speech sounds (for example, an /s/ substituted by a /t/), are assumed to be related to a cognitive-linguistic level. Phonetic processes, i.e., processes not resulting in loss of phonemic distinction (for example, when the Swedish dental fricative /s/ is realized as an interdental), are assumed to be related to articulatory problems. However, the view that certain types of speech errors would implicate the level of deficit has been questioned (Hewlett, 1990).

In the 1980s, the view of how children with cleft palate acquire their speech sound systems changed from focusing on physical mechanisms to also incorporating learning factors and strategies to compensate for the cleft (Moller, 1990). The level of the deficit of articulatory/phonological processes in children with cleft palate has been discussed. Some investigators have the opinion that deviant articulatory patterns in turn can cause unusual phonological patterns that persist even when the patophysiological conditions have been improved, for example after a successful palate repair (Grunwell and Russell, 1988; Chapman, 1993; Harding and Grunwell, 1996). The phonological patterns are then seen as a secondary phonological disorder resulting from a primary articulatory deviance. According to Hewlett (1985), however, persisting backing after a successful palate repair can be caused by incorrectly acquired motor patterns, and, therefore, does not necessarily imply a phonological disorder on the cognitive-linguistic level: "...a phonological substitution is one in which an incorrect phoneme is correctly realized; a phonetic distortion is one in which the correct phoneme is incorrectly realized (whether or not this phonetic realization crosses a phonemic boundary)" (p. 158). In this project, phonological analyses were performed to assess active processes in children with unilateral cleft palate, without taking a position on the level of the deficit.

When using measures of per cent correct consonants, no distinction between phonetic and phonological errors is applied. The measure percentage of consonants correct was originally developed by Shriberg and Kwiatkowski (1982). They measured the proportion of correctly articulated consonants in phonetic transcriptions of conversational speech in order to assess the "severity of involvement". Since then, several other investigators have applied this measure to report consonant accuracy (Chapman and Hardin, 1992; Morris and Ozanne, 2003; Chapman et al., 2008; Lohmander and Persson, 2008). In the measurement of per cent correct consonants, all errors are given the same weight, even if some errors may be age

appropriate. However, if the measure is adjusted for age, it instead indicates the degree of severity of articulatory and phonological errors at specific ages (Shriberg, 1993).

### 1.2.3 Narrative retelling

Production of narratives is regarded as an ecologically valid task for assessment of language skills in children (Paul and Smith, 1993; Botting, 2002). In narrative analysis, several language abilities used regularly in a child's educational environment may be assessed (Merritt and Liles, 1989). There are two types of narratives used in narrative analysis: story generation (i.e., spontaneous speech samples) and story retelling. In retelling, the narratives usually are longer and more complete with more complex story grammar than is the case in freely produced narratives (Merritt and Liles, 1989). Reproduction of narratives requires several well-functioning underlying cognitive abilities at a high level (Leinonen et al., 2000). The child needs to understand the task and the relationship between the input text and the topic. The input text should also be remembered and processed at the required speed, and, if there is picture support, visual and lexical information need to be integrated in the narrative (Leinonen et al., 2000). In addition, well functioning pragmatic skills are essential (Leinonen et al., 2000, Botting, 2002).

The Bus Story Test (Renfrew, 1997) is a standardized retelling test, which has been regularly used in primary language units in the UK (Botting, 2002). The test may predict persistent language impairment (Bishop and Edmundson, 1987) and also literacy performance in adolescence (Stothard et al., 1998). A translation of the Bus Story Test into Swedish has been published with reference data on the outcome measures, the information score, the mean length of utterance based on words, and the number of subordinate clauses, based on 100 Swedish-speaking children between the ages 3;9 to 6;8 years (Svensson and Tuominen-Eriksson, 2002).

### 1.2.4 Age appropriate development

Over the years, several theories related to children's phonological development have been presented. As early as 1941, Roman Jakobson presented ideas based on structuralist language



theory. Some aspects of this theory are considered adequate even today, for example, that the most extreme distinctions between open and closed articulation, which are universal, are the distinctions a child learns first, while the subtle distinctions, which are more specific to individual languages, are achieved later. Several decades later, other researchers stated that babbling may vary between individuals but in general follows universal patterns (e.g., Oller, 1980; Stark, 1980; Roug et al., 1989). Early consonant inventory in English-speaking children from 15 months to 24 months of age consists of oral plosives, nasals, and glides (Stoel-Gammon, 1985). Fricatives and liquids develop later. Anteriorly produced consonants (i.e., labials and alveolars) develop before posteriorly produced consonants (Stoel-Gammon, 1985).

Jakobson (1941) emphasized the distinction between phonetic-articulatory ability and the gradually developing ability of phonological organization and claimed there was no association between babbling and language development. In the 1980s, however, several researchers observed a continuity in the development from early consonant inventory in babbling to articulation and phonology in meaningful speech (Locke, 1983; Stoel-Gammon, 1985; Vihman and Greenlee, 1987). A theory that has been widely adopted in speech and language pathology is natural phonology (Stampe, 1979). According to this theory, there are phonological and natural simplification processes within a child, which facilitate speech production. These processes are gradually suppressed during development.

Phonological simplification processes displayed in children in pre-school years can be described as either syntagmatic or paradigmatic. Syntagmatic processes are context dependent, change the phonotactic and prosodic structures of a word, and are displayed in early years. An example of this is reduction of syllables as when /ba' na:n/ is realized as /na:n/ (English: banana). Most syntagmatic processes in Swedish-speaking children without cleft palate cease by the age of 3 years (Lohmander et al., 2014). The paradigmatic processes usually cease later than the syntagmatic processes (Lohmander et al., 2014). They are context independent, affect classes of segments, and do not change the structure of a word. An example is stopping, for example the fricative /s/ being replaced by the stop /t/ as when /su:l/ is realized as /tu:l/ (English: sun).

According to norm data of phonological/phonetic simplification processes in Swedish-speaking 3- and 5-year-olds (Lohmander et al., 2014), simplification of the voiceless fricative

/s/ was displayed in 73% of the 3-year-olds and in 32% of the 5-year-olds. Simplification of the voiceless fricative /ç/ was displayed in 46% of the children at 3 years of age and in 2% at 5 years of age. Simplification of the liquid /r/ was displayed in 59% at 3 years of age and in 17% at 5 years of age. Dentalization (e.g., the velar stop /k/ realized as the dental stop /t/) was displayed in 12% at 3 years of age and in 1% at 5 years of age. In addition, at 3 years of age, 7% displayed stopping, 6% voicing (e.g., the unvoiced stop /p/ realized as the voiced stop /b/), 3% devoicing, and 3% substitution of an oral consonant for /h/.

Usually, before 1 year of age, children start producing clauses consisting of one word, and, between the ages of 1;6 and 2;6, words are combined into a clause (Håkansson and Hansson, 2007). Between the ages 2;6 and 3, clauses get more extended and subordinate clauses are also used. At about 4 years of age, all types of subordinate clauses are established, together with simple and frequent grammatical constructions. Further on, grammatical complexity is developed (Håkansson and Hansson, 2007).

Grammatical ability does not develop in isolation but as part of overall language development along with phonological, lexical, and pragmatic development (Håkansson and Hansson, 2007). According to Leonard (1998), the ability to perceive, process and/or produce morphemes (the smallest meaningful units of language) with low degrees of phonetic substance, morphemes with shorter durations than the surrounding morphemes, is a prerequisite for age appropriate grammatical development. This means that, as is the case with other aspects of language, grammatical development is dependent on the ability to perceive and process auditive information.

There are indications of a close interaction between phonetic progress in babbling and lexical development (e.g., McCune and Vihman, 2001). The influences between the phonological system and vocabulary have been found to be bi-directional (e.g., Edwards et al., 2004). Up to about 18 months of age, the lexical development of children with age appropriate development is slow, and vocabulary contains few words and is not phonetically stable (Nettelbladt, 2007a). Thereafter, there is a substantial increase in the rate of vocabulary expansion, and, at about 2;5 years of age, the vocabulary contains about 500 words (Barrett, 1995). At 6 years of age, American English-speaking children have achieved about 14,000 words, and vocabulary continues to grow (Clark, 1995).

Conversational skill requires articulatory skill as well as ability in all linguistic domains (Nettelbladt, 2013). In addition, a child's contributions to a conversation need to occur at the right occasions with precision, and this presupposes interactional skills. Small children can already interact with a forthcoming adult at the pre-linguistic stage. Linguistic pragmatic development, however, assumes a certain level of language development. At the beginning, desires and opinions are expressed in rudimentary ways. Gradually, the ability to initiate and maintain conversations increases, and the child masters more complex and abstract topics of conversation (Nettelbladt, 2013). At 5 years of age, an age appropriate developing child can follow a conversational turn, predict its ending, and understand its intended meaning (McTear, 1985).

#### 1.2.5 Development in children with cleft palate

As in children without cleft palate, there is continuity in development in children born with cleft palate, from consonant production in babbling to articulation in meaningful speech (e.g., Chapman et al., 2003; Lohmander and Persson, 2008). Number of consonant types and frequency of occurrence of dental plosives at 18 months, for example, has been found to correlate significantly with per cent correct consonants at 3 years of age (Lohmander and Persson, 2008). Significant correlations have also been found between consonant production in babbling and mean length of utterance and lexical measures in meaningful speech (Chapman et al., 2003; Chapman, 2004).

Babbling in children born with cleft palate in general contains more nasal consonants and less pressure consonants than babbling in children without cleft palate (e.g., Chapman, 1991; Lohmander-Agerskov et al., 1994; Willadsen and Albrechtsen, 2006). In studies of children with un-operated cleft palate at the time of assessment, glottal sounds have dominated in babbling (e.g., Grunwell and Russell, 1987; Chapman, 1991; Chapman et al., 2001). On the other hand, in children with early closure of the soft palate but with an un-operated residual cleft in the hard palate at the time of assessment, the occurrence of glottal consonants has been low, as has that of anterior consonants. Instead, velar oral consonants, /k, g/, have been common (Lohmander-Agerskov et al., 1994; Lohmander et al., 2004; Willadsen and Albrechtsen, 2006).

About 50% of children born with cleft palate display impaired speech and/or phonology at 3 years of age (e.g., Chapman et al., 2008; Lohmander and Persson, 2008; Willadsen, 2012). Some phonological simplification processes have been described as specific to children with cleft palate. For example, backing (i.e., a dental or alveolar /t/ being produced posteriorly as the velar /k/) is common among children with cleft palate but unusual among children without cleft palate (Chapman and Hardin, 1992; Chapman, 1993; Harding and Grunwell, 1996). Nasal realization (i.e., an oral consonant such as /b/ may be realized as the nasal /m/) or nasal assimilation (i.e., the presence of nasal consonants in a word resulting in other consonants also becoming nasal) (Chapman and Hardin, 1992; Morris and Ozanne, 2003) and differences in voiced/voiceless stop production (Harding and Grunwell, 1996) are other processes described as cleft palate related in the literature. In addition, the phonology in children with cleft palate has been reported to be unstable and varying, with persisting immaturities and systematic sound preferences (Harding and Grunwell, 1996). At about 5 years of age, differences in phonological processes between children with and without cleft palate have been reported to decline (Chapman, 1993).

Glottal articulation/reinforcement, active nasal fricatives, and pharyngeal fricatives are other processes described as cleft palate related (Harding and Grunwell, 1998). When these processes occur in Swedish-speaking children, they are defined as active articulatory processes since the target phoneme is not perceived as other Swedish phonemes but rather as speech sounds that do not exist in Swedish.

Only two longitudinal studies on nasality in children with cleft palate have been published (Lohmander-Agerskov et al., 1998; Lohmander and Persson, 2008). According to the results, hypernasality and audible nasal air leakage subsided between 3 and 5 years of age, although 20 to 30 % of the children still had some degree of hypernasality at 5 years of age. In both these studies, the cleft in the hard palate was unrepaired at age 3 years and, in the first study, also at age 5 years.

Toddlers with cleft lip and palate have scored significantly lower on cognitive and linguistic measures than have peers without clefts (Jocelyn et al., 1996; Broen et al., 1998). Also, lexical ability and mean length of utterance have been found to be significantly poorer in toddlers with cleft palate when compared to peers without cleft (Scherer and D'Antonio, 1995). This may partly be explained by the phenomenon of lexical selectivity (i.e., individual

patterns of lexical selection and avoidance reflecting the production capability of a child), which has been observed in both children born with (Willadsen, 2013) and without cleft palate (Schwartz and Leonard, 1982). Although no significant differences have been found between children with and without cleft palate at pre-school or early school age regarding vocabulary (Collett et al., 2010a; Chapman, 2011) and grammatical skills (Chapman, 2011), there are indications that a rather high proportion of children born with cleft palate perform more poorly on standardized tasks of expressive grammar and vocabulary (Young et al., 2010).

Speech difficulties may also be related to pragmatic skills (Frederickson et al., 2006). A passive conversational style (i.e., responding to initiatives by a conversational partner but rarely initiating conversational turns) has been found to be more common among children with cleft palate, compared with peers without cleft palate. Possible explanations for this passive conversational style may be a true pragmatic deficit, shy personality, or poor speech intelligibility (Frederickson et al., 2006). Such factors may lead to unwillingness to speak

Although individuals with clefts is a heterogeneous group and many children perform within normal limits in the areas of speech and language, school-aged and adolescent children may have difficulties in several linguistic areas, including rapid verbal labelling, verbal fluency, and verbal memory (Conrad et al., 2009). There also are also indications of impaired reading skills among individuals with cleft lip and palate (Collett et al., 2010b; Chapman, 2011; Conrad et al., 2014).

#### 1.2.6 Variables that may influence outcomes in children with cleft palate

Factors other than the cleft palate itself may affect speech and language development in children with cleft palate, such as methods for primary palatal surgery. However, despite extensive research on speech outcomes after primary palatal surgery, there is no evidence regarding which surgical method gives the best speech outcome. Factors complicating the evaluation of cleft palate intervention are the multidimensionality of outcomes, the duration of follow-up, the reproducibility and validity of outcome measures, the diversity of management, and small sample sizes (Roberts et al., 1991). Further, the possibility of comparing different treatment procedures reliably has been limited due to a lack of

standardized methods for collection and analysis of speech data (Lohmander and Olsson, 2004; Sell, 2005). The impact of the chosen speech material on speech judgement when assessing cleft palate speech has not been studied previously.

According to a review of articles on speech outcome after primary palatal surgery in individuals with unilateral cleft lip and palate, the most common surgical procedures were the Wardill-Kilner pushback closure, the Van Langenbeck technique, and a two-stage procedure with delayed hard palate closure (Lohmander, 2011). No significant differences were found in speech outcomes related to one-stage surgery with the Van Langenbeck technique or the Wardill-Kilner pushback closure (Pigott et al., 2002; Farzaneh et al., 2008), and contradicting results were reported when the Furlow procedure was compared to the Van Langenbeck technique (Spauwen et al., 1992; Van Lierde et al., 2004). Speech outcome in two-stage procedures has been reported to be at least as good as speech outcome after one-stage procedures (e.g., Van Demark et al., 1989; Lohmander et al., 2006; Lohmander et al., 2012).

In recent decades, interest in prospective evaluations of speech outcomes after standardized surgical procedures for cleft palate repair has increased. One example is the Scandicleft project, a randomized clinical trial in which cleft palate centres from Sweden, Norway, Denmark, Finland, and the UK participated (Semb et al., 2013). Children with complete unilateral cleft lip and palate were included and randomized to treatment according to each centre's ordinary method or another method, implying soft palate closure at 3 to 4 months of age and hard palate closure at 12 months of age. Preliminary results at age 5 years revealed significantly higher numbers of retracted/backed oral consonants in children treated with soft palate closure at 3 to 4 months of age and hard palate closure at 36 months of age, compared to children treated with soft palate closure at 3 to 4 months of age and hard palate closure at 12 months of age. Further, children treated with hard palate closure at 3 to 4 months of age and soft palate closure at 12 months of age had significantly higher total numbers of non-oral consonants than did children treated with soft palate closure at 3 to 4 months of age and hard palate closure at 12 months of age (Willadsen et al., 2013).

Another important variable is hearing. Since hearing loss decreases a child's access to speech, there is a hypothesized negative relationship between otitis media with effusion and the development of speech and language (Roberts et al., 2004). The incidence of otitis media with effusion and related mild to moderate hearing loss is higher among children

born with cleft palate than it is in peers without cleft palate (Flynn et al., 2009). Children born with cleft palate often have problems with dilating and opening the Eustachian tube due to impaired muscle function (Arnold et al., 2005) and a hypercompliant Eustachian tube (Sheahan and Blayney, 2003). The dysfunction results in difficulties in equalizing pressure and in draining secretions in the middle ear, and, thus, negative pressure and tympanic membrane retractions.

However, the relationship between hearing impairment and speech and language development in children with cleft palate is unclear. For example, in one study there was a significant correlation between mild hearing impairment and consonant inventory at 12 months of age, but at 18 months of age this correlation was no longer significant (Lohmander et al., 2011). In another study, hearing status at 12 months of age correlated with both scores of comprehension and expressive language at 24 months of age (Jocelyn et al., 1996).

There are also other explanatory models for linguistic and cognitive problems among individuals with clefts, for example, models related to neurobiological aspects (e.g., Ceponiene et al., 1999; Goldsberry et al., 2006; Shriver et al., 2006); however, these aspects were not covered in the present project.

### **1.3 RATIONALE FOR THE INCLUDED STUDIES**

Approximately 50% of the children born with cleft palate present speech difficulties around 3 years of age. Published studies point towards a relationship between early limitations of articulation/phonology and limitations in other abilities of expressive language. A longitudinal perspective is, therefore, warranted. Several factors can influence the results when evaluating cleft palate speech and phonology. The impact of the speech material chosen for assessment is one such factor. The method of surgery for primary palate repair is another. However, the influence is unclear and needs to be further investigated. Furthermore, expressive language, including phonology, has been sparsely explored in children with cleft lip and palate and extended knowledge is essential in order to improve the intervention for this patient group.

## **2 AIMS**

### **2.1 GENERAL AIMS**

The general aims of this project were to:

- Assess the effectiveness of different speech materials used when evaluating cleft palate speech.
- Assess longitudinal development of speech and expressive language in children born with unilateral cleft lip and palate compared with that of children without cleft.
- Identify variables in babbling and early speech, which may be associated with later expressive language in children born with cleft palate.

### **2.2 SPECIFIC AIMS**

The specific aims of each study were to:

- Study I: Clarify differences in speech outcome related to different speech materials in 5-year-olds with and without cleft palate and to estimate the reliability and validity of the speech materials.
- Study II: Elucidate phonological development in Swedish-speaking 3-year-olds born with unilateral cleft lip and palate compared with that in peers without cleft, and to see if any measures of oral consonant production at 18 months might be associated with phonological skill at 3 years of age.
- Study III: Clarify if there are any differences in speech and phonology in 3-year-olds born with unilateral complete cleft lip and palate treated with three different surgical methods.
- Study IV: Elucidate phonological ability in Swedish-speaking 5-year-olds born with unilateral cleft lip and palate compared to that in their peers without cleft, and to clarify the relationship with performances at 3 years of age.
- Study V: Investigate expressive language skills in terms of narrative competence in retelling in 5-year-olds with unilateral cleft lip and palate, and to explore if there is a relationship between these language skills and articulatory and phonological ability at ages 3 and 5 years.



### 3 MATERIAL AND METHODS

#### 3.1 PARTICIPANTS

##### 3.1.1 Distribution of participants in different groups and studies

A total of 50 children were included in the project, 30 children with unilateral cleft lip and palate and a comparison group of 20 children without cleft lip and palate. All children were monolingual Swedish-speaking and without any known additional malformations or syndromes. They were distributed in four groups:

- Eleven children (five girls and six boys) with unilateral cleft lip and palate, born between 1997 and 2003, treated with a two-stage closure with soft palate closure between 3.4 and 6.4 months and hard palate closure at a mean age of 12.3 months (two-stage 12).
- Nine children (four girls and five boys) with unilateral cleft lip and palate, born between 1997 and 2003, treated with a two-stage closure with soft palate closure between 3.4 and 6.4 months and hard palate closure at a mean age of 36.2 months (two-stage 36).
- Ten children (three girls and seven boys) with unilateral cleft lip and palate, born between 2005 and 2008, treated with a one-stage closure at a mean age of 13.6 months (one-stage).
- Twenty children (11 girls and nine boys) without cleft lip and palate, born in 2000 (comparison group). According to a parental questionnaire, their development was considered to be age-appropriate, including in terms of language development.

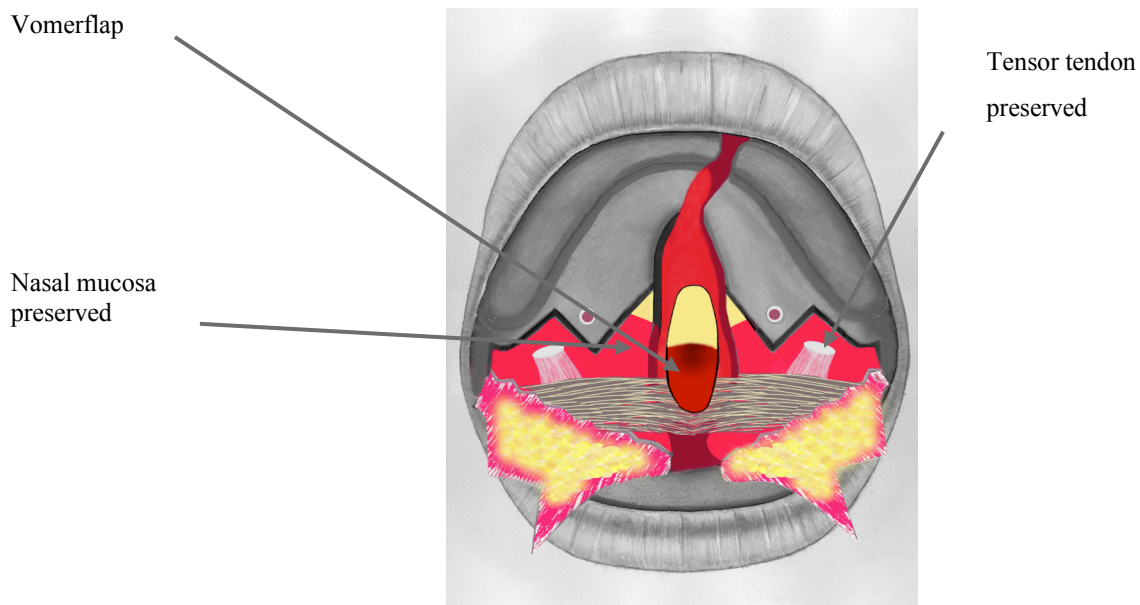
The number of children from different groups participating in the five studies is presented in Table 1.

*Table 1.* The number of children participating in the different studies.

<b>Group</b>	<b>Study I</b>	<b>Study II</b>	<b>Study III</b>	<b>Study IV</b>	<b>Study V</b>
Two-stage 12	11	9	9	11	11
Two-stage 36	9	9	9	9	9
One-stage	0	0	10	9	9
Comparison group	20	20	0	20	20
Total	40	38	28	49	49

### 3.1.2 Surgical methods

Two surgeons at Sahlgrenska University Hospital treated the 20 consecutive children with unilateral cleft lip and palate from the western region of Sweden between the years 1997 and 2004. The technique used was early soft palate repair with delayed hard palate closure at the age of 12 or 36 months according to the Scandcleft protocol (Friede et al., 2013) (Figures 6 and 7).



*Figure 6.* Soft palate repair according to the Scandcleft procedure. Reprinted with permission from Jan Lilja.

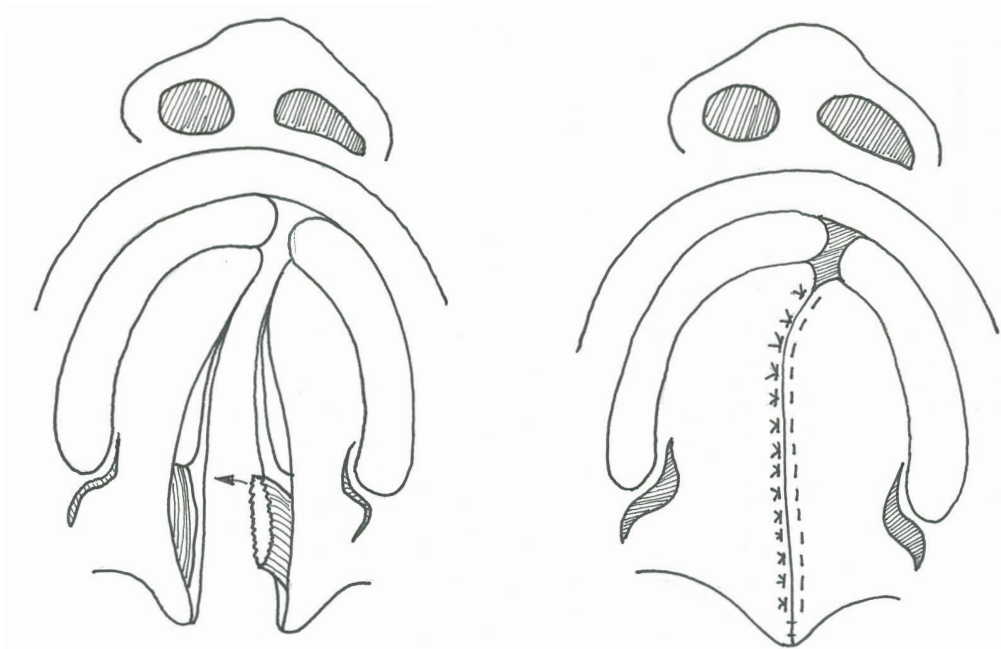


*Figure 7.* To the left, a residual cleft in the hard palate after soft palate repair according to the Scandcleft procedure. To the right, a child treated with primary palatal closure in two stages according to Scandcleft procedure. A residual cleft in the alveolar ridge is left un-operated to be closed in the mixed dentition at 8 to 9 years of age. Printed with kind permission from parents.

In this procedure, a zigzag incision is made at the border between the soft and hard palate, which continues anteriorly on the palatal shelf. After incisions have been made along the cleft at the border between the nasal and oral mucosa backwards to the uvula, a small mucoperiosteal flap is raised. This flap includes, anteriorly, mucoperiosteum and, posteriorly, the mucosa of the oral layer, which is dissected from the muscles. A vomer flap, which is posteriorly and cranially based, is then raised from behind the vomero-premaxillary suture. The vomer flap is turned over and sutured into the anterior half of the nasal layer. This facilitates the closure of the soft palate and will also anchor it to the vomer. However, the nasal layer is not detached from the posterior part of the hard palate. An intra-velar veloplasty is performed wherein the muscles are dissected from the nasal mucosa without attention to the tensor tendon. The muscles are then repositioned posteriorly, sutured in the midline, and covered with the mucosal part of the oral flaps. The mucoperiosteal part of the flaps covers the raw surface of the vomer flap. The cleft in the hard palate is closed on a later occasion in one layer, using a cranially based vomer flap. Nineteen children underwent this procedure. For one child, a two-layer-closure with mucoperiosteal flaps was used.

The 10 consecutive children from the southern region were treated by one surgeon at Skåne University Hospital between the years 2005 and 2009 with intra-velar veloplasty, according to the method developed by Sommerlad (2003) (Figures 8 and 9). The surgery was performed under 3.5 loop magnification. The procedure used is as follows. Incisions are made along the cleft on both sides at the border of the oral and nasal mucosa. Mucoperiosteal flaps are then raised, particularly exposing the posterior border of the hard palate. The greater palatine neurovascular bundle is released from its foramen to facilitate closure of the oral layer. With a stay suture closing the uvula, meticulous muscle preparation can start. The levator muscle and the tensor tendon are separated from the posterior hard palate, and the tensor tendon can be bisected medial to the hamulus in order to release tension. The musculature is then further separated both from the oral and nasal mucosa. Closure starts with the nasal layer after complementary release within the hard palate. Thereafter, the levator muscles are retropositioned and united in the midline, constituting the reconstruction of the levator sling. This dissection encompasses also the palatoglossus and the palatopharyngeus muscular fibres, although the levator is considered the important one. Finally, the oral layer is closed. In summary, the method comprises minimal hard palate dissection with radical retropositioning of the velar musculature and tensor tenotomy. In case tension of the closure is perceived, the

procedure can be supplemented with releasing lateral incisions, and this option was utilized in four of the 10 children included in this study.



*Figure 8.* Primary palatal closure in one stage, with intra-velar veloplasty according to Sommerlad (2003), in combination with lateral releasing incisions. Illustration by Liisi Raud Westberg.



*Figure 9.* A child treated with primary palatal closure in one stage with intra-velar veloplasty according to Sommerlad (2003). A residual cleft in the alveolar ridge is left un-operated to be closed in the mixed dentition at 8 to 9 years of age. Printed with kind permission from parents.

### 3.1.3 Hearing

Hearing ability was assessed with audiometry by a paediatric audiologist on the same day as each child's speech and language were assessed, and is presented in Table 2. The children affected with hearing loss had mild hearing loss (21–40 dB).

*Table 2.* Per cent of children with hearing data with hearing loss (21–40 dB) at the ages of 18 months, 3 years, and 5 years.

<b>Groups</b>	<b>Per cent with hearing loss</b>		
	<b>18 months</b>	<b>3 years</b>	<b>5 years</b>
Two-stage 12	44	36	18
Two-stage 36	67	56	33
One-stage	No data	30	22
Comparison group	20	10	15

### 3.1.4 Speech-language therapy

No child with unilateral cleft lip and palate had received speech-language therapy before 3 years of age. Between the ages of 3 and 5 years, eight children received two or three sessions of speech therapy, four children seven to 10 sessions, and one child 25 sessions. The remaining 16 children did not receive any speech therapy (Table 3).

*Table 3.* Number of received sessions of speech-language therapy in the three groups of children with unilateral cleft lip and palate treated with different methods for primary palatal surgery between the ages of 3 and 5 years.

<b>Number of sessions</b>	<b>Two-stage 12 (n=11)</b>	<b>Two-stage 36 (n=9)</b>	<b>One-stage (n=9)</b>
0	5	3	8
2-3	5	3	0
7-10	1	2	1
25	0	1	0

### 3.1.5 Parents' educational background

The parents of children with unilateral cleft lip and palate from the western region and of children without cleft lip and palate reported their educational backgrounds using a questionnaire. There was a significant difference (Mann-Whitney U test) between the two groups, with higher educational levels in the parents' of children without cleft palate (mothers' educational background  $Z = -3.734$ ,  $p = <.001$ ; fathers' educational background  $Z = -3.096$ ;  $p = .002$ ).

## 3.2 SPEECH SAMPLES USED FOR ANALYSIS

All children were audio and video recorded at 3 and 5 years of age. In addition, the children with unilateral cleft lip and palate from the western region (the two-stage 12 and two-stage 36 groups) and the comparison group were audio and video recorded at 18 months of age. Speech samples analysed at different ages are presented in Table 4.

*Table 4.* Speech samples analysed at the different ages.

<b>18 months</b> <b>(One-stage not included)</b>	<b>3 years</b>	<b>5 years</b>
Utterances from 45–60 minutes babbling	Single word naming test	Single word naming test
		Sentence repetition
		The Bus Story Test
		About 2 minutes of conversational speech

### 3.2.1 Speech samples

*Babbling:* At 18 months of age, a fixed set of age-appropriate toys was used for elicitation of babbling during interaction with a parent and a speech-language pathologist (Lohmander et al., 2011). A median number of 98 (range 34–100) consecutive speech-like utterances (i.e., involving at least one vowel-like or consonant-like element or consonant-vowel combinations) were used.

*Single word naming:* A single word test by picture naming, developed in the Scandleft Project to assess the production of consonants vulnerable to a cleft condition (i.e., oral stops and fricatives) (Lohmander et al., 2009) was used. It consisted of 32 pictures (including two dummies) at 3 years of age and 33 pictures for eliciting single words at 5 years of age. Thirty pictures/words were the same at both ages.

*Sentence repetition:* The children repeated 13 short sentences after the test leader. The sentences contained different high-pressure consonants, low-pressure consonants, nasal consonants, or mixed consonants.

*The Bus Story Test:* The test is a narrative task where the children are asked to retell the story with the aid of 12 pictures (Renfrew, 1997; Svensson and Tuominen-Eriksson, 2002).

*Conversational speech:* At 5 years of age, the test leader asked the children about their daily life or talked about pictures or a jigsaw puzzle.

### 3.2.2 Recording and editing

Speech was documented with audio recordings and simultaneous video recordings. In study III, video files were used for analysis, and, in the other studies, audio files were used. All children were recorded in a room at Sahlgrenska University Hospital or at Skåne University Hospital. For the children at Sahlgrenska University Hospital, speech was documented with digital audio recordings (Sony Walkman TCD-D8; Sony Corp., New York) using a condenser microphone (Sony ECM-MS957) and a high quality video camcorder with an external microphone (Sony ECM-MS957). Audio files from the video recordings were used for analysis of the children recorded at Skåne University Hospital in studies IV and V. The equipment used at Skåne University Hospital was a video camera (Canon HF10) with an external microphone (Sony ECM-M5957).

The recordings were transferred to .wav-files for editing in Adobe Audition 2.0 or Audacity. The recordings at 18 months of age used in study II had been prepared and used in a previous study (Lohmander et al., 2011). For analysis at 3 years of age in study II, un-edited recordings were used. In the other studies, the recordings were edited in separate files for word naming, sentence repetition, retelling of the Bus Story, and conversational speech. In studies I, II, and V, the recordings were prepared for blinded analysis.

### **3.3 ASSESSMENT**

#### **3.3.1 Phonetic transcription**

At 18 months of age, already completed phonetic transcriptions from a published study of babbling were available (Lohmander et al., 2011). At 3 and 5 years of age, narrow phonetic transcription was performed by the author according to the IPA and ExtIPA conventions (IPA, 2002; IPA, 2005). In studies I, II, IV, and V, about 30% of the recordings, randomly selected, were re-transcribed by the author and a second independent transcriber for reliability assessment. In study III, 100% of the recordings were re-transcribed by the author and a second independent external transcriber for the same purpose.

#### **3.3.2 Rating of passive cleft speech characteristics**

In study III, the passive cleft speech characteristics hypernasality, hyponasality, and audible nasal air leakage were each rated by the author on an ordinal scale from 0 to 3, where 0 means normal resonance and no audible nasal air leakage and 3 means severe deviation of resonance and audible nasal air leakage occurring always or almost always. For the purpose of intra-rater reliability assessment, all recordings were re-rated by the author after one month. In addition, a second independent judge re-rated all recordings for assessment of inter-rater reliability.

### **3.4 ANALYSIS**

#### **3.4.1 Oral consonants at 18 months of age**

In study II, each child's stable consonants were assessed from the transcriptions at 18 months of age. A consonant had to appear on at least three occasions to be regarded as stable (Chapman, 1991; Willadsen and Albrechtsen, 2006). The total number of oral consonants, oral stops, anterior oral stops, and dental/alveolar oral stops as well as the number of different oral consonants and oral stops were examined for correlation with the primary outcomes at 3 years of age (per cent correct consonants adjusted for age, number of established phonemes, total number of phonological processes).



### 3.4.2 Outcomes of articulation and phonology at 3 and 5 years of age

Outcomes of articulation and phonology, for comparison between materials and groups, or for correlation analysis, are presented in Table 5. In study I, per cent correct consonants, per cent correct places, and per cent correct manners (Lohmander and Persson, 2008) were calculated. In the other studies, the measure per cent correct consonants adjusted for age was used instead of per cent correct consonants. Per cent correct consonants adjusted for age was based on the same scoring rules for calculation as per cent correct consonants. It was, however, modified with respect to age-appropriate articulatory and phonological simplification processes, i.e., varying types of lisp and weakening of /r/ were scored as correct.

*Table 5.* Measures of phonology and articulation at 3 and 5 years of age used for comparisons between speech materials (M) or groups of children (G), or for correlation analysis (C) in the different studies.

Measures	Study I	Study II	Study III	Study IV	Study V
Per cent correct consonants	M				
Per cent correct places	M				
Per cent correct manners	M				
Per cent active cleft speech characteristics	M		G		
Per cent phonological simplification processes	M				
Per cent correct consonants adjusted for age		G/C	G	G/C	C
Number of established phonemes		G/C			
Total number of phonological processes		G/C	G		
Number of different phonological processes		G	G		
Number of different syntagmatic processes		G			
Number of different paradigmatic processes		G			
Number of consistent phonological processes				G	

In study II the number of established phonemes (i.e., phonemes correctly produced in at least 50% of the possible instances) was assessed (Lohmander et al., 2014). Analyses of phonological processes were performed in all four studies. In study I, per cent phonological simplification processes was calculated as a quotient by dividing the number of phonological simplification processes by the total number of consonants of the sample and multiplying it by 100. In studies II and III the total number and number of different phonological processes were assessed for each child. In addition, in study II, the number of different syntagmatic and paradigmatic phonological processes was calculated. In study IV, the number of consistent phonological processes was assessed. Processes occurring in at least 20% of all possible occurrences were judged as consistent (McReynolds and Elbert, 1981). The outcome per cent active cleft speech characteristics (i.e., retracted oral articulation, glottal stops and glottal reinforcement, /h/ used for oral consonants, nasal realization of voiceless fricatives, nasal realization of other oral consonants, pharyngeal fricatives) was used in studies I and III. Additionally, in studies II, III, and IV, descriptive analyses of phonological and articulatory processes were performed, including active cleft palate-related processes.

### 3.4.3 Analysis of narratives

In study V, the recordings of the Bus Story Test were orthographically transcribed and assessed according to the test manual (Renfrew, 1997; Svensson and Tuominen-Eriksson, 2002). The information score, the mean length of utterances based on words, and the number of subordinate clauses were calculated for each child from the orthographic transcriptions. After two months, about 30% of the recordings, randomly chosen, were re-transcribed and re-assessed by the main assessor (the author) and an independent assessor. The results of the Bus Story Test were compared between groups and also correlated with per cent correct consonants adjusted for age at 3 and 5 years of age.

### 3.4.4 Reliability testing

In study I, reliability was an outcome, and is, therefore, presented in the result section. Reliability in the other studies, by means of inter- and intra-rater agreement, is presented in Table 6. At 18 months, consonant transcriptions from a published study where reliability was presented were used for analysis (Lohmander et al., 2011).

*Table 6.* Reliability in studies II–V, by means of inter- and intra-rater agreement, in the group with unilateral cleft lip and palate (UCLP), the comparison group without cleft (COMP), and in all children.

<b>Measure evaluated</b>	<b>Method for agreement Calculation</b>	<b>Inter UCLP</b>	<b>Inter COMP</b>	<b>Inter all</b>	<b>Intra UCLP</b>	<b>Intra COMP</b>	<b>Intra all</b>
Phonetic transcription from audio recordings at 18 months; babbling*	Mean agreement** of place and manner of articulation, 20% of the material	> 80%	> 80%	> 80%	> 90%	> 90%	> 90%
Phonetic transcription from audio recordings at 3 years; word test	Median agreement** of consonants, 30% of the material	70%	86%	78%	88%	95%	92%
Phonetic transcription of video recordings at 3 years; word test	Mean agreement** of consonants, 100% of the material	77%	-	-	90%	-	-
Phonetic transcription of audio recordings, 5 years; word test	Median agreement** of consonants, 30% of the material	91%	91%	91%	98%	99%	99%
Phonetic transcription of audio recordings, 5 years; Bus Story Test	Median agreement** of consonants, 30% of the material	85%	86%	86%	91%	97%	97%
Hypenascality on a 4-point scale	Mean agreement, one scale value difference accepted, 100% of the material	71%	-	-	100%	-	-
Hyponasality on a 4-point scale	Mean agreement, one scale value difference accepted, 100% of the material	100%	-	-	100%	-	-
Audible nasal air leakage on a 4-point scale	Mean agreement, one scale value difference accepted, 100% of the material	96%	-	-	100%	-	-
Information score (Bus Story Test)	Intraclass correlation coefficient, 30% of the material	-	-	0.969	-	-	0.989
Mean length of utterance (Bus Story Test)	Intraclass correlation coefficient, 30% of the material	-	-	0.973	-	-	0.969
Number of subordinate clauses (Bus Story Test)	Intraclass correlation coefficient, 30% of the material	-	-	0.863	-	-	0.959

\* From (Lohmander et al., 2011)

\*\* All mean and median agreement values of consonant transcriptions were calculated point by point (Bialocerkowski and Bragge, 2008)

### 3.5 STATISTICAL ANALYSES

Statistical analyses were made under the supervision of a professional statistician. Median and range values were used for descriptive analyses, and, in study V, mean values and standard deviations were also presented. Nonparametric statistics were used due to the small group sizes and skewed distributions of data. Since there are no alternatives in non-parametric statistics, a two-way ANOVA was performed to control for possible variables that may influence the outcomes in study II. In study III, first the Kruskal-Wallis test was used, and then a post hoc pair wise analysis, using the Mann-Whitney U test with Bonferroni correction, was performed for variables that displayed significant differences. For all statistical analyses  $p < 0.05$  (two-tailed) was considered to indicate significant results. The tests used for statistical analysis are presented in Table 7.

Table 7. Tests used for statistical analysis in the different studies.

Test	Study I	Study II	Study III	Study IV	Study V
Wilcoxon matched pair signed rank test	X				
Mann-Whitney U test		x	x	x	x
Two-way ANOVA		x			
Kruskal-Wallis test			x		x
Spearman's rank order correlation test		x		x	x

### 3.6 ETHICAL APPROVALS

The Regional Research Ethics Committee of Gothenburg (R257-97) approved participation of the children with unilateral cleft lip and palate from the western region, and the Regional Ethical Review Board of Lund (D-nr: 548/2008) approved the enrolment of the children from the southern region. All parents gave written informed consent for participation.

## **4 RESULTS**

### **4.1 STUDY I**

Differences in per cent speech accuracy in different speech materials and reliability of speech materials by means of inter- and intra-transcriber agreement of consonant transcriptions were assessed in 5-year-olds with and without cleft palate. The medians of intra- and inter-transcriber agreement were good (varying between 79.5 and 98.9%) in both groups and all sampling modes. The children with cleft palate displayed significantly higher per cent correct consonants and less active cleft speech characteristics in word naming than in all other sampling modes. They also displayed higher per cent correct places in word naming than in sentence repetition and conversational speech. Additionally, they achieved better results regarding per cent correct manners and per cent phonological simplification processes in word naming than in conversational speech. Children without cleft palate achieved good results, irrespective of sampling mode.

### **4.2 STUDY II**

The phonology in Swedish-speaking children born with unilateral cleft lip and palate at age 3 years was compared with the phonology in peers born without cleft. In addition, measures of oral consonant production at 18 months, which may be associated with phonology at 3 years of age, were explored. At 3 years of age, the group with unilateral cleft lip and palate displayed significantly lower per cent correct consonants adjusted for age, a lower number of established phonemes, and a higher total number of phonological processes compared with the group without cleft. The significant differences persisted after adjustment for parental educational background. The descriptive analysis revealed both characteristics related to the cleft palate and phonological processes seen in children with age-appropriate development at an earlier age in the group with unilateral cleft lip and palate. A varying phonology was also found to be more frequent among the children with unilateral cleft lip and palate than among the peers without cleft. Variables at 18 months correlating significantly with per cent correct consonants adjusted for age at 3 years in the group with unilateral cleft lip and palate were: total number of oral consonants, oral stops, dental/alveolar oral stops, and number of different oral stops.

### **4.3 STUDY III**

Articulation, passive cleft speech characteristics, and phonology at 3 years of age in children with unilateral complete cleft lip and palate treated with three different methods for primary palatal surgery were assessed. The group treated with a one-stage closure at about 13 months of age showed significantly better results regarding per cent active cleft speech characteristics and total number of phonological processes than did the children in the group treated with a two-stage surgery who still had an un-operated hard palate. There also was a significant difference in hypernasality; however, due to low inter-rater agreement, the results on hypernasality were not reliable. There were no significant differences between outcomes of children treated with a two-stage surgery and hard palate closure at 12 months of age and outcomes of the two other groups treated with other methods for primary palatal surgery.

### **4.4 STUDY IV**

The phonology at age 5 years in children with unilateral cleft lip and palate compared to that of peers without cleft palate and the relationship with performances at 3 years of age was assessed. The group with unilateral cleft lip and palate displayed significantly lower per cent correct consonants adjusted for age and higher number consistent phonological processes at age 5 years than did peers without cleft. However, the results among the children with unilateral cleft lip and palate varied widely. The correlations between the outcomes at ages 5 and 3 years were strong. No relationship was found between speech difficulties and the number of speech-language therapy sessions when the children were reviewed individually. The results indicated poorer phonology in the children treated with a two-stage surgery with hard palate closure at age 3 years than in the children treated with palate repair at an earlier age.

### **4.5 STUDY V**

Information score, mean length of utterance, and number of subordinate clauses when retelling the Bus Story were assessed in children with and without unilateral cleft lip and palate. No significant differences between the groups were found. However, there was a strong trend towards significantly lower results on the information score among the children

with unilateral cleft lip and palate compared with the comparison group. This trend was not related to differences in the surgical method for primary palatal repair or to gender. Furthermore, 65.5% of the children in the group with unilateral cleft lip and palate had an information score below 1 standard deviation from the norm value, compared with 30% in the comparison group. Nine children in the group with unilateral cleft lip and palate and two children in the comparison group scored 2 standard deviations below the mean norm value. No relationship was found between the outcomes of the Bus Story Test and the number of speech-language therapy sessions when the children were reviewed individually. In the children with unilateral cleft lip and palate, there was no significant association between the results of the Bus Story Test and articulatory and phonological competence, neither at the same age nor earlier.

## **5 DISCUSSION**

In this doctoral project, speech and expressive language in children with unilateral cleft lip and palate were longitudinally explored. In addition, the effectiveness of different speech materials used when assessing cleft palate speech was evaluated.

### **5.1 LONGITUDINAL DEVELOPMENT OF SPEECH AND EXPRESSIVE LANGUAGE**

Children with cleft palate are a heterogeneous group. Even when studying a group of children with a specific type of cleft palate, for example non-syndromic children with unilateral cleft lip and palate as in this project, the outcomes of different variables vary widely. This indicates that there may be sub-groups among the carefully included children with different prerequisites for speech and language development (Morris and Ozanne, 2003).

According to the results of this project and in agreement with other studies (e.g., Chapman, 1993; Lohmander and Persson, 2008; Collett et al., 2010a; Young et al., 2010; Chapman, 2011), some children show considerable difficulties with speech and language at ages 3 and 5 years whereas other children of the same ages have speech and language skills comparable to those of typically developing peers. Children with speech and language problems at these ages may be in need of speech and language intervention in order to prevent problems at school age. This is of great importance both for the individual as well as from a social and academic perspective. Although many individuals with cleft lip and/or palate are successful at school, as a group, children with clefts do not succeed equally well in school when compared with peers without clefts. For example, significant deficits in educational achievement in compulsory school were found among adolescents born with cleft lip and/or palate in a population-based Swedish register study (Persson et al., 2012). Educational achievement may be hampered by poor reading skills. Further, children with cleft lip and palate have been found to score significantly lower than control groups on knowledge and use of letters at about 5 to 7 years of age, and better scores on early reading measures were associated with better speech (Chapman, 2011). As a group, children with non-syndromic orofacial clefts have also been found to score significantly lower than control groups at 5 to 7 years of age on



basic reading, phonological memory, and reading fluency (Collett et al., 2010b). Hence, it is important that children who are at risk for prolonged problems with language, reading, and writing are identified early, and that the correct action is taken to prevent learning problems at school.

From this perspective, the connections in this project between the outcomes at 18 months and 3 years of age and between 3 years and 5 years of age are highly interesting. Measures of consonant production in babbling were associated with per cent correct consonants adjusted for age at 3 years of age (II), which is in agreement with earlier findings (Chapman et al., 2003; Lohmander and Persson, 2008). Also, there was a significant correlation between per cent correct consonants adjusted for age at ages 3 and 5 years (IV), consistent with the findings of Lohmander and Persson (2008). In addition, a significant relationship between per cent correct consonants adjusted for age at 3 years of age and number of consistent phonological processes at 5 years of age was found (IV), and this further strengthens the connections between earlier and later outcomes.

In speech analysis, the main focus was on active processes, i.e., phonological simplification processes and active cleft speech characteristics. The prevalence of backing in the group with unilateral cleft lip and palate was high at both 3 and 5 years of age (II, III, IV), which is in accordance with findings of a high prevalence of retracted oral articulation in a previous study on Swedish-speaking children with unilateral cleft lip and palate (Lohmander and Persson, 2008). Both characteristics related to the cleft palate and phonological processes seen in typically developing children at an earlier age have previously been found in English-speaking (Chapman, 1993; Harding and Grunwell, 1996) and Danish-speaking children (Willadsen, 2012), and these findings were verified in the present project (II, III, IV).

Even at 5 years of age, many of the children with unilateral cleft lip and palate had impaired phonology (IV). This is an important finding. Previously, at most 10 children with cleft palate have been included in investigations of phonology in 5-year-olds with cleft palate, and no significant differences compared to children without cleft palate were found (Chapman, 1993). Given the indications of a connection between speech and early reading skills in children with cleft palate (Chapman, 2011), phonological problems at this age should be treated.

In study III passive cleft speech characteristics were also assessed. About a third of the children with unilateral cleft lip and palate had moderate to severe hypernasality at 3 years of age. This is a better outcome than in the studies by Lohmander and colleagues (Lohmander-Agerskov et al., 1998; Lohmander and Persson, 2008) where all children had an un-operated residual cleft in the hard palate at age 3 years, which probably influenced the results. The results on hypernasality, however, were not entirely reliable in the present project. This is discussed below. In total, few studies have evaluated speech after primary palatal surgery at about age 3 years (Spauwen et al., 1992; Zanzi et al., 2002; Chapman et al., 2008), and only one has reported measures on hypernasality (Spauwen et al., 1992). In that study, 50% of the children treated with Furlow palatoplasty were reported to have hypernasal speech, as compared to no such occurrence in children treated with the van Langenbeck procedure; however, no measures of reliability were reported.

Regarding the ability to retell information, a larger proportion of children with unilateral cleft lip and palate than without had problems at 5 years of age (V). There was, however, no association between retelling ability and articulatory/phonological skills. One variable that influences the ability to retell information is pragmatic skill. Pre-schoolers with cleft lip and palate have been found to be less conversationally assertive compared to peers without cleft lip and palate (Frederickson et al., 2006). Other skills, such as rapid verbal labelling, verbal fluency, and verbal memory that influence the ability to retell information, and are related to underlying cognitive abilities and language processing, may be impaired in individuals with non-syndromic orofacial clefts (Conrad et al., 2009). These aspects have not been investigated among pre-school children with cleft palate and should be further explored.

## **5.2 IMPACT OF SURGERY, HEARING, AND OTHER POTENTIALLY INFLUENTIAL VARIABLES**

In accordance with other studies (Willadsen, 2012; Willadsen et al., 2013), the findings indicate that it is not favourable to have the hard palate repaired as late as at 3 years of age, at least when performed according to the surgical procedure studied (III). Even at 5 years of age, the effect of an un-repaired hard palate at 3 years of age was noticeable (IV). Other variables related to surgery that may affect speech outcome, although rarely discussed, are surgical skill and caseload, in that a higher number of palatal repairs undertaken by a surgeon

is associated with better speech outcomes (Williams et al., 1999). This project was not designed to elucidate the impact of surgical experience, however, as all three surgeons were very well trained in cleft palate surgery.

In previous studies, children treated with an early closure of the soft palate, even with an open cleft in the hard palate at the time of assessment, have displayed a proportionately high occurrence of oral stops in their babbling (e.g., Willadsen and Albrechtsen, 2006; Lohmander et al., 2011; Willadsen, 2012), whereas non-oral and low-pressure consonants have been common in the babbling of children with an un-operated palate at the time of assessment (e.g., Chapman et al., 2001; Scherer et al., 2008). There are indications of a continuity in the development from consonant production in babbling before surgery to consonant production in connected speech after surgery (Chapman et al., 2003) and also of a relationship between the occurrence of oral stops at 18 months and per cent correct consonants at 3 years of age (Lohmander and Persson, 2008). Thus, it could be assumed that the children in the present project treated with a one-stage palate closure at about 13 months of age would have poorer scores of per cent correct consonants adjusted for age than the children treated with early soft palate closure at 3 years of age. This was not the case, however. It would have been interesting to assess the occurrence of oral stops in babbling and the continuity to later consonant production in the children treated with a one-stage closure also, but, as babbling in these children had not been documented, this was unfortunately not possible.

Since otitis media with effusion and related hearing loss is very common in children born with cleft palate (Flynn et al., 2009) and hearing loss can potentially influence speech and language development, hearing loss was controlled for when studying phonological ability at 3 years of age (II) and also when studying expressive language at 5 years of age (V). At 3 years of age, differences between children with and without unilateral cleft lip and palate in per cent correct consonants adjusted for age, number of established phonemes, and total number of phonological processes persisted after analysing the possible effect of hearing (II). No correlations were found between hearing and expressive language in narrative retelling at 5 years of age (V). Thus, according to the results in the present project, hearing loss did not influence outcomes of phonological ability and narrative retelling. One reason for this might be that hearing loss in most children with unilateral cleft lip and palate in the present project was minimal (unilateral and mild). Also, hearing loss related to otitis media with effusion is fluctuating and needs to be assessed regularly and frequently in order to provide a reliable

picture of a child's hearing history. According to findings by Flynn and Lohmander (2014), abnormal middle ear status decreased and hearing across frequencies improved in children with unilateral cleft lip and palate up to 5 years of age. Thereafter, however, the children with unilateral cleft lip and palate still had a higher prevalence of abnormal middle ear status compared with peers without cleft, and hearing in the high frequencies did not significantly improve. Abnormal middle ear status and impaired hearing in the high frequencies may lead to challenges in speech production, auditory processing, and academic achievement, hence the effect of hearing loss on different speech and language variables needs to be further investigated (Flynn and Lohmander, 2014).

Socio-economic status may influence the language of toddlers; however, the effect may vary in different cultures (Berglund et al., 2005). In a Swedish study, socio-economic status did not affect communicative skills at 18 months of age (Berglund et al., 2005). In the present project, the parents answered questions about educational background as a measure of socio-economic status (II). There was a significant difference between children with and without cleft lip and palate in terms of the higher educational levels of the parents of children without cleft palate. However, the significant differences between all primary outcomes at 3 years of age of children born with and without unilateral cleft lip and palate persisted after adjustment for parental educational background. This is in accordance with the findings in a Danish study where phonological development at 3 years of age was unrelated to the educational level of the parents (Willadsen, 2012).

Speech-language therapy should at best have a positive impact on speech and language. Of the 29 participating children with unilateral cleft lip and palate at age 5 years, 13 had received therapy from a speech-language pathologist between the ages of 3 and 5 years. Only five children had received therapy for more than three sessions. When the children were reviewed individually, no pattern regarding the relationship between speech and language difficulties and the number of speech-language therapy sessions could be discerned, and the number of therapy sessions a child attended seemed not to have been related to the degree of speech and language difficulties (IV, V). Children with cleft palate and speech and language difficulties would probably benefit from speech-language therapy. To date, however, there is little evidence supporting any specific method for speech-language therapy in children with cleft palate (Bessell et al., 2013).

## 5.3 METHODOLOGICAL ISSUES

### 5.3.1 Participants

Small group size is a common limitation in studies evaluating speech outcome after surgical repair (Roberts et al., 1991). A total of 30 children with unilateral cleft lip and palate were included in this project, which is a rather small group, and the subgroups related to different methods for primary palatal surgery were very small. Individual differences, thereby, had a large impact on the results. For example, the children in one sub-group may have had innately better phonological abilities, not related to method for palatal repair, than was the case with the children in another sub-group, although the children in the subgroups were consecutively selected. Another fact that needs to be considered is that the children in the two subgroups operated on in two stages were born between 1997 and 2003, and the children in the subgroup operated on in one stage were born between 2005 and 2008. It cannot be excluded that a general improvement of intervention over time favoured the outcomes in children treated with the one-stage procedure.

No formal genetic or cognitive tests were performed on the participants included in the project, but children with known additional malformations or syndromes were excluded since other problems might influence outcomes, particularly when comparing small groups. One child with unilateral cleft lip and palate was later diagnosed with attention deficit hyperactivity disorder. Such a disorder may affect the results of the Bus Story Test (Miniscalco et al., 2007). When comparisons between children with and without unilateral cleft lip and palate were re-calculated with this child excluded, the trend towards a difference between groups actually decreased (from  $p = 0.051$  to  $p = 0.065$ ). It is possible that more children with undiagnosed additional problems were included in the group, which may have influenced the results.

Not only do small study groups constitute a limitation in clinical research, but the fact that documentation is not always complete also does so. At 3 years of age, the recordings of two children treated with two-stage palatal closure with hard palate closure at 12 months were missing. These two children were included in the phonological analyses at 5 years of age. In addition, at 5 years of age, the recording of one child treated with one-stage closure was missing. This child was included in the analysis at 3 years of age. When the samples are as

small as in the present project, inclusion or exclusion of an individual child might significantly affect the results.

### 5.3.2 Ethics

The assessments were performed in association with ordinary routine follow-ups conducted by the cleft palate teams. Participation in the project resulted in some extra assessments for the children with unilateral cleft lip and palate, which might have been tiresome for some of them. Ethical review had approved participation of the children with unilateral cleft lip and palate, and all parents had given their written informed consent. However, the children themselves had not been asked if they wanted to participate. Doing so would have been more ethically correct (De Lourdes Levy et al., 2003). In study V, for example, three children with unilateral cleft lip and palate and two without declared that they did not want to participate in the retelling task. They were enticed by the test leader to retell as much as possible of the Bus Story, which resulted in information scores of 2 standard deviations below the mean norm value. When these children were excluded from the statistical analysis, no significant difference in information score between children with and without unilateral cleft lip and palate was found. The issue of the assent of the participating children, however, may be considered a dilemma. Since not wanting to participate may be related to linguistic difficulties, the exclusion of un-willing children could bias the results.

### 5.3.3 Speech data

Audio files and video files were available for all analyses. The audio files were chosen for analysis in all studies including both children with and without cleft lip and palate. The reason for this was to make the listening un-biased regarding which children were diagnosed with unilateral cleft lip and palate and which children were not. Visual articulatory cues may get lost when using audio recordings instead of video recordings (Sell, 2005); however, un-biased listening was valued higher in these analyses.

The issue of unintelligible speech samples is a methodological problem rarely discussed in literature. Spontaneous conversational speech has long been recommended for assessment of disordered speech (Grunwell et al., 1993). At 3 years of age, samples of both single word

naming and conversational speech were available. It would have been interesting to perform phonological analysis on coherent speech at 3 years of age. This would have provided more occurrences of syntagmatic processes. Many children, however, had extensive sequences of unintelligible conversational speech at 3 years of age, which had to be excluded, resulting in samples too small for analysis. Therefore, the samples of single word naming were chosen for analysis (II, III). The advantage of this was also that the samples were standardized. At 5 years of age (IV), single word naming was chosen for analysis to make comparisons with the outcomes at 3 years of age possible. In addition, retelling of the Bus Story was chosen for phonological analysis of coherent speech (Renfrew, 1997). Still, at age 5 years, some children had unintelligible coherent speech; however, in most cases, the target consonants were known to the assessor in the Bus Story samples.

Although sentence repetition was not used for phonological analysis of connected speech in study IV in this project, sentence repetition is a speech material reflecting coherent speech with high reliability and validity (I) and has been found useful when assessing speech in older children with persistent speech impairments (Howard, 2013). When using sentence repetition, speech material is standardized and the assessor knows the target phonemes. In addition, it is easy to administer in that it is not as time-consuming to elicit and analyse, as is the case with retelling and conversational speech.

#### 5.3.4 Perceptual assessment and analysis

When comparing different methods of treatment, it is essential that listening is performed blinded and within the same time frame to avoid listener bias. Blind consensus listening has been recommended to ensure a consistency of assessors (Mehendale and Sommerlad, 2003). This was, however, not practical within this project. In study III, the main listener knew the children recorded at Skåne University Hospital. In order to control that this did not influence the outcomes, a second listener re-transcribed and re-rated all recordings. Inter-transcriber and inter-rater agreement was considered good in most cases. However, mean inter-rater agreement was poor for hypernasality, which made the results unreliable. Low intra- and inter-rater agreement in the assessment of hypernasality has been reported in many other studies (e.g., Karling et al., 1993; Keuning et al., 1999; Timmons et al., 2001; Lohmander and Persson, 2008) and may be solved by systematic and frequent training (Lee et al., 2009; Sell et

al., 2009). In study III, calibration of the raters was performed, but systematic and frequent training was not accomplished. This turned out to be insufficient for the rating of hypernasality.

In study IV, the main transcriber transcribed the samples of the 5-year-old children recorded at Sahlgrenska University Hospital blinded. The samples of the children recorded at Skåne University Hospital were transcribed un-blinded at a later occasion. Since it cannot be excluded that this could have affected the results, no statistical comparisons between the sub-groups treated with different methods for primary palatal surgery were performed.

Calculation of per cent correct consonants was performed in study I. In the following studies, per cent correct consonants adjusted for age were used since this was considered a more interesting measure as it relates to age appropriate development (Shriberg, 1993). The focus was merely on active processes, phonological as well as articulatory; thus, passive cleft speech characteristics were not scored as incorrect. In the published norm data of Swedish-speaking children, passive characteristics were also scored as incorrect (although with only a few occurrences), and no adjustment for age was considered (Lohmander et al., 2014). Hence, it is important to consider differences in methodology when results from different studies are interpreted and compared.

### 5.3.5 Assessment of expressive language

In this project, phonological analysis was performed using phonetic transcriptions. A limitation of phonetic transcriptions is that speech sounds are evaluated with reference to discrete categories, which may make the transcription too simplistic and lead to fine-grained differences in the quality of speech sounds, i.e., covert contrasts not being described (Strömbergsson, 2014). Strömbergsson (2014) found that “clear substitutions” of [t] for /k/ and [k] for /t/ in children with a phonological disorder were rated as less prototypical than correct productions when listeners were allowed to use a visual-analogue scale. This was also found in a small study on cleft palate speech (Eriksson and Ferm, 2000). Further, English-speaking listeners have been found to have difficulties perceiving palatal stops (a phonemic category that does not exist in English) in cleft palate speech (Santelmann et al., 1999). Regarding children with cleft palate in this project who displayed oral backing as a frequent



phonological process, a /t/ maybe was not always substituted by a /k/ but rather produced as a consonant sound in between /t/ and /k/, meaning there actually was an attempt to signal the phonological contrast between the two. This information may have been missed in the stage of phonetic transcription in studies II, III, and IV. Thus, even if phonetic transcription actually does allow usage of symbols for unusual pronunciation such as “in between /t/ and /k/”, a listener might have difficulties deciding on the place of articulation and choosing a symbol. Further, in cases where the “in between production” actually was transcribed as a palatal oral plosive, it was incorporated in the category palatal/velar/uvular in the stage of analysis. This means that, with a more careful analysis, these details might have been taken account of.

The phonological processes were quantified in order to be included in the statistical analyses. Compared with calculation of per cent correct consonants, where consonant production is scored as correct or not correct based upon agreed guidelines (Shriberg and Kwiatkowski, 1982), analysis of phonological simplification processes is more qualitative in nature as it depends on how a judge chooses to categorize different processes. An illustrative example is consonant deletion, which may be judged as one category or be divided into three: initial deletion, medial deletion, and final deletion. Notwithstanding this fact, it was considered valuable to include quantitative measures of phonological processes in the statistical analyses since the focus was on phonology in several studies.

In the current project, analyses of phonological simplification processes were performed without taking a position on the level of the deficit. It would have been interesting to also study the relationship between meta-phonological competence and articulatory/phonological processes (Bird et al., 1995) in order to increase the knowledge of the underlying causes of phonological problems in children with cleft palate. However, this was not possible since no data on meta-phonological ability had been retrieved.

According to the general aims of this project, an intention was to assess longitudinal development not only of articulation/phonology but also of other aspects of expressive language in children born with unilateral cleft lip and palate as compared with children without cleft palate. At 3 years of age, we set out to assess vocabulary and mean length of utterance based on the samples of conversational speech. However, substantial parts of the

samples of conversational speech at 3 years of age were unintelligible. Thus, it was not possible to perform further linguistic analyses at 3 years of age.

At 5 year of age, expressive language in retelling was assessed. The advantage of using the Bus Story Test for assessment is that norm data has been published for Swedish regarding information score, mean length of utterances based on words, and the number of subordinate clauses (Renfrew, 1997; Svensson and Tuominen-Eriksson, 2002). In addition, the Bus Story Test was used for assessment of phonology (Renfrew, 1997). Analysis of story grammar was also considered in the present project (e.g., Stein and Glenn, 1979); however, since the Bus Story does not meet the criteria for a story, story grammar was not analysed. According to Ochs and Taylor (1992), a narrative includes a central incident and following reactions whereas a report only has to consist of events in a sequence. Pursuant to this definition, the Bus Story is a report rather than a narrative. Other analyses, also assessing pragmatic aspects, would have been interesting to perform (Holck et al., 2011), but this was not practical within the frames of the current project.

### 5.3.6 Statistical analysis

Due to small groups sizes and skewed distribution of data, mainly non-parametric statistical analyses were performed. In addition, in study II, a two-way ANOVA was used to control for hearing and parental education level, variables, which potentially could influence the outcomes. This was questioned by two of the reviewers. One of them suggested that the children with unilateral cleft lip and palate with normal hearing, the children with unilateral cleft lip and palate with hearing loss, the children without cleft lip and palate with normal hearing, and the children without cleft lip and palate with hearing loss should constitute separate subgroups in the statistical analysis. However, this would have resulted in very small groups for analysis. Since there is no alternative to two-way ANOVA in non-parametric statistics, and the alternative had been to refrain from controlling for hearing and parental education level, it was decided to keep the two-way ANOVA.

In study III, differences among sub-groups treated with different methods for primary surgical repair were tested with the Kruskal-Wallis test. For variables displaying significant differences, a post-hoc analysis with the Mann-Whitney U test with Bonferroni correction ( $p$

$<.05 = <.017$ ) was used. Bonferroni correction is used when multiple tests are performed, such as comparing more than two groups, assuming that a finding depends on the number of the other tests performed. The use of Bonferroni correction has been questioned. For example, according to Perneger (1998), adjusting statistical significance for the number of tests that have been performed on study data creates more problems than it solves in that it increases the likelihood of type II errors so that truly important differences are deemed non-significant. In study III, however, the use of Bonferroni correction did not change the results regarding significant differences between groups.

## 6 CONCLUSIONS AND CLINICAL IMPLICATIONS

The results strongly indicate a high prevalence of deviant phonology among Swedish-speaking 3- and 5-year-olds born with unilateral cleft lip and palate. Further, obvious difficulties retelling information at 5 years of age were found in the group and these were unrelated to articulatory and phonological ability at ages 3 and 5 years. A wide variation in outcomes of expressive language among children with unilateral cleft lip and palate was verified, clearly suggesting a sub-group with expressive language deficits. Articulatory and phonological skills at 3 and 5 years of age may partly be related to surgical methods for primary palatal repair. A two-stage palatal procedure with hard palate closure as late as 3 years of age, using the surgical technique in the present project, may be disadvantageous for the development of speech and phonology compared with surgical procedures where the entire palate is closed at an earlier age, and should be avoided.

An important reflection from the findings is that speech and language pathology resources need to be concentrated on the children at risk for future speech and language problems. Measures of oral consonants and oral stops at 18 months of age were associated with phonology at 3 years of age in children with unilateral cleft lip and palate, and may be useful for identifying children in need of further speech-language intervention. In addition, there was a strong relationship between consonant production at 3 years of age and phonology at 5 years of age. Poor consonant production at age 3 years will, thus, signal a risk for poor phonology at age 5 years, and attention should be paid to this.

Finally, when assessing the best speech performance in children with cleft palate, word naming is the most reliable and valid sampling mode. If the purpose is to assess coherent speech, sentence repetition is recommended since it is a reliable and valid speech material and is also easy to administer.

## **7 FUTURE STUDIES**

The results of the present project need to be verified in larger groups of children and in children with other types of clefts. Children with additional problems should also be included. The use of covert contrasts among Swedish-speaking children with cleft palate should preferably be investigated in order to improve knowledge of phonological development in this group. This would contribute to the development of methods for phonological intervention in children born with cleft palate. Also, the causes of problems retelling information in children with cleft palate needs to be explored, for example by assessing pragmatic skills and abilities related to language processing. Furthermore, studies on language and reading skills in children with cleft palate at school age and the association of this with skills of expressive language and meta-phonology at pre-school age are highly warranted in order to find children at risk for reading difficulties and persisting language impairments and to achieve the possible prevention of such problems.

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