Internationally adopted children with unilateral cleft lip and palate

– longitudinal perspectives on speech production and language ability

AnnaKarin Larsson

Speech and Language Pathology Unit
Department of Health and Rehabilitation
Institute of Neuroscience and Physiology
Sahlgrenska Academy
University of Gothenburg

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UNIVERSITY OF GOTHENBURG

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annakarin.larsson@vgregion.se

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To Alvar, Calle and Anders

Abstract

Many children with cleft lip and palate have been adopted to Sweden. Most had had no palatal closure performed in their native countries and received their first palatal surgery in Sweden while acquiring a new first language.

The main aim of this thesis was to investigate speech production (i.e. consonant proficiency, consonant errors and velopharyngeal competence) and language ability at school age in internationally adopted children from China with unilateral cleft lip and palate. The thesis included data based on phonetic transcriptions and perceptual ratings of audio-recorded standardised routine speech assessments analysed by blinded raters (Studies I–III), standardised speech and language assessments (Study IV) and parental ratings (Studies III and IV).

In *Study I*, speech production was investigated in 14 children at age 3 years and compared with a group of non-adopted children with the same cleft type. Results showed that the internationally adopted children performed significantly lower than the non-adopted children on consonant proficiency and on perceived velopharyngeal competence. In *Study II*, 25 internationally adopted children were assessed at age 5 years and compared with non-adopted children with the same cleft type. Results showed that the internationally adopted children had significantly fewer correct consonants and more restricted consonant inventories than the non-adopted children. A high proportion (52%) had an incompetent velopharyngeal function, although they did not differ significantly from their non-adopted peers. *Study III* longitudinally investigated speech production in 17 children between the ages of 3 and 7–8 years. Additionally, relationships between speech production and expressive language at age 7–8 years were studied. Significant progress in consonant proficiency and velopharyngeal competence from age 3 years onwards was found. However, at age 7–8 years more than 80% of the children had a consonant proficiency score at least 2 SD below the age-specific norms. Additionally, the children exhibited difficulties on measures of expressive language. In *Study IV*, receptive and expressive language ability was investigated in 27 internationally adopted children at age 7–8 years. Comparisons were made with a group of internationally adopted children without cleft lip and palate. The only variable that significantly differed between groups was speech ability, where the children with cleft lip and palate scored lower according to both tests and parental ratings. However, both groups scored low on expressive language ability compared with test norms.

In conclusion, internationally adopted children with cleft lip and palate develop their speech-production ability considerably despite having later palatal repair than non-adopted peers. However, many still have speech difficulties at school age. Many of the internationally adopted children, regardless of whether they had a cleft lip and palate, presented with poor expressive language ability at age 7–8 years, and a risk of delayed language development many years after adoption was found in many children.

Keywords

Internationally adopted, cleft lip and palate, speech production, language ability

Sammanfattning på svenska

Under 2000-talet skedde flera förändringar vad gäller internationella adoptioner. Fler barn med särskilda behov än någonsin tidigare adopterades till Europa och Sverige. Många barn var födda med läpp-käk-gomspalt och de flesta hade inte fått någon kirurgisk slutning av gommen innan adoption. För att kunna tillägna sig ett funktionellt tal krävs en hel gom och möjlighet att sluta/separera näshåla och munhåla från varandra. De adopterade barnen fick oftast sina första insatser från sjukvården samtidigt som de började lära sig ett nytt förstaspråk.

Det övergripande syftet med föreliggande avhandling var att studera talproduktionen (konsonantproduktion, konsonantfel och gomfunktion) hos internationellt adopterade barn från Kina med unilateral läpp-käk-gomspalt i ett longitudinellt perspektiv samt att studera deras språkliga förmåga i tidig skolålder. Avhandlingen bestod av fyra delstudier av barn i tre åldersgrupper: 3, 5 och 7–8 år. Data till studierna bestod av analyser av fonetiska transkriptioner och perceptuella lyssnarbedömningar av ljudinspelat talmaterial från standardiserade rutinundersökningar av tal som gjorts av läpp-käk-gomspaltsteamet vid Sahlgrenska universitetssjukhuset i Göteborg (delstudie I–III), en standardiserad tal- och språkbedömning vid 7–8 år (delstudie IV) och föräldraskattningar (delstudie III–IV). Journaldata har också inhämtats (delstudie I–III).

I *delstudie I* undersöktes talproduktionen hos 14 barn vid 3 års ålder och jämfördes med en grupp icke-adopterade barn med samma spalttyp. Resultaten visade att de adopterade barnen presterade signifikant sämre än icke-adopterade barn på alla talproduktionsmått. I *delstudie II* undersöktes talproduktionen hos 25 adopterade barn vid 5 års ålder. Jämförelser gjordes med icke-adopterade barn i samma ålder och med samma spalttyp. Resultaten visade att de adopterade barnen hade signifikant sämre konsonantproduktion och mindre konsonantinventorium än de icke-adopterade barnen. Många (52 %) bedömdes också ha inkompetent gomfunktion, men någon signifikant skillnad mellan grupperna förelåg inte. *Delstudie III* var en longitudinell studie av talproduktionen mellan 3 och 7–8 års ålder hos 17 barn. Sambandet mellan talproduktion och expressiv språkförmåga i skolåldern undersöktes också. Resultaten visade på stor utveckling gällande talproduktion från 3 års ålder och framåt, men det framkom även att majoriteten av barnen hade kvarstående talsvårigheter vid 7–8 års ålder. Dessutom framkom svårigheter gällande expressiv språklig förmåga vid den åldern. I *delstudie IV* undersöktes receptiv och expressiv språklig förmåga hos 27 adopterade barn med unilateral läpp-käk-gomspalt. Jämförelser gjordes med en grupp adopterade barn utan spalt samt med testnormer. Resultaten visade att endast talförmågan skilde sig åt mellan grupperna; barnen med läpp-käk-gomspalt presterade signifikant lägre. Detta gällde både utifrån testning och utifrån föräldraskattningen. Däremot visade resultaten att båda grupperna presterade lågt gällande expressiv språklig förmåga jämfört med testnormer.

Sammanfattningsvis har avhandlingen visat att internationellt adopterade barn med läpp-käk-gomspalt utvecklar sin talproduktion mycket från 3 års ålder trots att de får senare gomslutning än icke-adopterade barn men att många hade kvarstående talsvårigheter i skolåldern. När det gäller barnens språk tycks läpp-käk-gomspalt i sig inte öka risken för försenad språkutveckling. Många av de adopterade barnen, oavsett om de hade läpp-käk-gomspalt, presterade lågt gällande expressiv språklig förmåga och tycks vara i riskzonen för att ha försenad språkutveckling många år efter adoptionen.

List of papers

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

I. Larsson, AK., Schölin, J., Mark, H., Jönsson, R., Persson, C.

Speech production in 3-year-old internationally adopted children with unilateral cleft lip and palate

International Journal of Language and Communication Disorders, 2017; 52:626–636.

II. Larsson, AK., Miniscalco, C., Mark, H., Schölin, J., Jönsson, R., Persson, C.

Internationally adopted children with unilateral cleft lip and palate – consonant proficiency and perceived velopharyngeal competence at the age of 5

Cleft Palate Craniofacial Journal 2020; 17 Jan. E-pub ahead of print.

III. Larsson, AK., Miniscalco, C., Mark, H., Jönsson, R., Persson, C.

Persisting speech errors and poor expressive language ability at school age – a longitudinal study of internationally adopted children with cleft lip and palate

Manuscript.

IV. Larsson, AK., Persson, C., Klintö, K., Miniscalco, C.

Internationally adopted children with and without a cleft lip and palate showed no differences in language ability at school-age

Acta Paediatrica 2020; 11 April. E-pub ahead of print.

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Abbreviations

BST	The Bus Story Test
CCC-2	Children's Communication Checklist, second edition
CELF-4	Clinical Evaluation of Language Fundamentals, fourth version
CLP	Cleft lip and palate
CSC	Cleft speech characteristic
DLD	Developmental language disorder
DSC	Developmental speech characteristic
HPC	Hard-palate closure
IA	Internationally adopted
MLU	Mean length of utterance
NA	Non-adopted
NGO	Non-governmental organisation
OME	Otitis media with effusion
PC	Palatal closure
PCC	Percent consonants correct
PCC-A	Percent consonants correct – adjusted for age
PCM	Percent correct manner of articulation
PCP	Percent correct place of articulation
PTA	Pure-tone average
SPC	Soft-palate closure
SLP	Speech-language pathologist
SVANTE	Swedish Articulation and Nasality Test
SVP	Secondary velopharyngeal
TROG-2	Test for Reception of Grammar, second version
UCLP	Unilateral cleft lip and palate
VP	Velopharyngeal
VPC	Velopharyngeal competence
VPI	Velopharyngeal insufficiency

Note. The abbreviations listed here are all abbreviations used in the running text. Some tables contain additional abbreviations, which are listed and explained below each table.

Introduction

For a period starting around 2008, cleft lip and palate (CLP) teams in Sweden were faced with a sudden large increase in the number of children with an unoperated CLP who had been adopted from other countries, mainly from China. Those children had been adopted at an older age than previously seen, and most of them had open palates.

While there was already a considerable amount of prior research into the speech and language development of internationally adopted (IA) children, as well as into that of non-adopted children with CLP, there was very limited knowledge when it came to IA children with CLP. In addition, the previous research involving IA children had focused in most part on children adopted at younger ages (around 1 year).

This is the background to the present project, which also evolved from everyday clinical practice at the Sahlgrenska University Hospital, Gothenburg, Sweden, and in close contact with the CLP team there. It should be noted that CLP is a 'special need' in the world of international adoptions, and prospective adoptive parents are usually advised to seek more knowledge about the specific special need(s) that they would consider before deciding to adopt a child with a special need. As the wave of adoptions of children with CLP was building up, Swedish CLP teams were thus contacted by prospective adoptive parents asking about the types and extent of interventions that their children would need. Importantly, many of them had questions regarding prognosis and wanted to know what to expect when it came to speech outcome for their children.

Hence the present PhD project was initiated to fill a void in clinical knowledge by making more knowledge available to clinicians and parents about the speech and language development of IA children with CLP.

International adoptions

International adoptions of children started after the Second World War, at a time when many orphans were left at institutions (Lindblad, 2004). Sweden experienced its first peak of international adoptions in 1970–1980, when South Korea and India were common countries of origin (Ballard et al., 2015; Children Above All Adoptions, 2020; Lindblad, 2004).

Starting in the early 1990s, there was a considerable increase in the number of international adoptions. International adoptions were formally authorised under Chinese law in 1992, and that represents the starting point for adoptions from China (Selman, 2017). It has been estimated that in 1992–2015, at least 145,000 children were adopted from China. This makes China the second-largest sending country in the world, after South Korea (Selman, 2017).

Since 2004, considerable changes have taken place regarding both countries of origin and receiving countries (Selman, 2015). Around that time, China became the top sending country of children for international adoptions, both globally and in relation to Sweden. However, after that year international adoptions from all countries started to decline (Selman, 2015), and those from China in particular. Possible causes mentioned in the literature include increases in domestic adoptions and decreases in the number of orphans (Ballard et al., 2015). What is more, the characteristics of the children adopted also changed. One major change which happened between 2005 and 2009 was a large increase in children with special needs adopted from China (Ballard et al., 2015). In 2005, only 9% of the children adopted from China had a special need, but in 2009, 49% of adoptions from China were special-needs adoptions. Corresponding changes were seen in all receiving countries, but Sweden was the country receiving the highest proportion of children with special needs from China in 2009. Having represented only 6% in 2005, special-needs adoptions accounted for 69% of all adoptions from China to Sweden in 2009 (Selman, 2015). To this should be added that there has been a change in the children's age at adoption. According to statistics from a Swedish adoption organisation, Children Above All Adoptions (2020), most children adopted prior to 1990 were below 1 year of age and only around 5% were above 2 years at the time of adoption. By contrast, in 2002–2012, more than 50% were above 2 years at the time of adoption, and in 2013–2019 over 70% were above 2 years. This is true at the European level as well: according to statistics pertaining to the Hague Adoption Convention, the proportion of children aged 5 or more at the time of adoption increased by a factor of almost ten between 2005 and 2009 (Selman, 2015). Finally, a shift was also seen in the sex ratio of adopted children. According to statistics for 1980–2019 from Children Above All Adoptions (2020), many more girls than boys were internationally adopted to Sweden in 1980–2001, but since 2001 the proportion of boys has been increasing, and boys now make up the majority.

Today, almost all international adoptions to Sweden are 'special-needs adoptions' (Adoptionscentrum, 2020; Children Above All Adoptions, 2020). It should be kept in mind that the term 'special needs' is problematic in that its definition varies greatly between and within authorities and adoption organisations in sending and receiving countries as well as, presumably as a consequence, in research. A special-needs adoption may be an adoption of a child who is older than 3 years, an adoption of several siblings at the same time and/or an adoption of a child with a known (or a suspected) medical diagnosis. The nature of such known or suspected medical diagnoses differs immensely. Examples of conditions that have been considered, in various contexts, to represent a special need include frequent infections, prematurity, hearing loss, skin conditions, congenital heart defects, craniofacial conditions, and suspected alcohol and/or drug exposure (Adoptionscentrum, 2020; Miller et al., 2016). However, the Guide to Good Practice of the Hague Adoption Conference (2008) defines a special-needs adoption as an adoption of a child who may be suffering from a behavioural disorder or a trauma, who is physically or mentally disabled, who is older (usually above 7 years) or who is part of a sibling group.

In summary, the world of international adoptions has changed a great deal over the past 15–20 years. In the past 10 years, an increasing number of children with special needs have been internationally adopted to the Western world (Raffety, 2019; Selman, 2015). Among those children, CLP is a common medical condition. A Swedish study from 2012 found that international adoptions to Sweden of children with CLP had become more common and that the mean age at adoption was around 2 years (Hansson et al., 2012). A majority of the children concerned had open palates on arrival and had undergone no palatal surgery in their country of origin.

Speech and language development in internationally adopted children

Children who are adopted to a different country usually change their language in conjunction with the adoption. The loss of their birth or native language is usually immediate (Glennen & Masters, 2002; Snedeker et al., 2012), and so is the start of their second process of first-language acquisition (De Geer, 1992). However, some studies question whether the birth language of international adoptees is fully 'lost', as remnants of the native language have been found to exist in IA adults decades after the adoption (Choi et al., 2017a Choi et al., 2017b; Hyltenstam et al., 2009; Pierce et al., 2014).

Language acquisition in IA children without CLP is widely studied, and there is a large body of evidence that children who change languages during the first two years of their life usually catch up with their non-adopted (NA) peers within 2–3 years in terms of proficiency in their new language (Glennen, 2014; Glennen & Masters, 2002; Roberts et al., 2005). Studies of language development in children adopted at ages above 2–3 years also report those children to be at language levels corresponding to the average for their age and/or in line with the levels of their NA peers (Glennen, 2014).

Previous research, which has usually studied language within the first two or three years after the adoption, has shown variable results in terms of the effect on language outcome exerted by the age at adoption, ranging from no association at all or just a small one to moderate or strong associations (Croft et al., 2007; Dalen, 2001; Glennen & Masters, 2002; Scott et al., 2011). Additionally, the potential effect of age at adoption on language outcome also differs between different language areas (Delcenserie, 2016; Scott & Roberts, 2016). Children adopted before the age of 2 years usually perform within average on tests of speech and language performance until they reach school age (Gauthier & Genesee, 2011; Roberts et al., 2005). However, at school age, there is some evidence of lower performance in IA children on language tests that involve executive functions (Desmarais et al., 2012; Eigsti et al., 2011) and more complex expressive grammatical tasks (Delcenserie et al., 2013; Gauthier et al., 2012; Scott et al., 2011).

Studies of language development in children adopted after the age of 2 years are more scarce, probably owing to the characteristics of children adopted internationally in the past – at the time when most studies were performed, the absolute majority of children were adopted at an early age. However, the few existing studies of children adopted at the age of 2 to 4 years found that they performed less well on language tests than their peers who had been adopted at a younger age (Glennen, 2009).

Another important issue when considering age at adoption is that children learn language at different rates depending on their age. On the one hand, a child who is older when adopted has more language to learn in order to catch up with NA peers than a younger child, but, on the other hand, such an older child in all likelihood also has a more developed cognitive ability and hence can learn at a faster rate. This was indeed shown by some researchers who found that children adopted before the age of 16 months learned new words at a slower rate than children adopted after the age of 30 months (Snedeker et al., 2012). A further factor that must be kept in mind when considering the potential impact of age at adoption is the impact of years spent at an institution or in other forms of living arrangements that do not represent a physically and/or psychologically favourable environment for an orphaned child. Institutionalised children are at risk of many different health and developmental problems (Miller & Hendrie, 2000). For example, although institutions and alternative living arrangements for orphaned children differ a great deal within and between countries, they do impose a heightened risk of delayed language development and other developmental difficulties (Loman et al., 2013; Miller & Hendrie, 2000; van Ijzendoorn et al., 2005).

A comprehensive study by Glennen (2014) investigated a group of 56 children on five occasions during their first three years post-adoption using a large battery of standardised assessments of articulation as well as receptive and expressive language.

Glennen also collected samples of language during play sessions to analyse the mean length of utterances (MLU). The children she studied had been adopted between the ages of 12 months and 4 years and 11 months. Receptive language reached expected levels relative to test norms faster than expressive language did. This was true for all children regardless of their age at adoption. However, children adopted at the age of 3 or 4 years needed more time to catch up and reach expected levels of receptive language than younger children, and improvements were still noted in their third year post-adoption. Similar results were found for expressive language, where the older children (adopted at 3 or 4 years of age) also needed more time to reach the levels expected for their age. It was also found that the children's MLU scores based on spontaneous-language samples were lagging behind other language areas assessed using a standardised language test (CELF). Regardless of their age at adoption, the children scored in the lower average range for MLU, and MLU was considered a 'significant area of weakness when compared with other expressive language skills 3 years after adoption' (Glennen, 2014, p. 199). By contrast, articulation proficiency was found to be the area which was easiest for all children to acquire, regardless of their age at adoption: two years post-adoption, all groups had reached age-expected levels in articulation proficiency.

When children are old enough to start school, the demands placed on their language capacity are higher. For optimal learning outcomes at school, children need to be able to process and produce more complex language. There is little research in this field, but some studies describe phonology and expressive and receptive vocabulary abilities in school-age IA children to be within average compared with test norms (Delcenserie, 2016). However, there are also studies describing that such children have difficulties in vocabulary and in expressive and receptive grammar compared with same-age NA peers (Delcenserie & Genesee, 2014; Delcenserie et al., 2013). Here it should be noted that few studies have performed detailed analyses of morphosyntactic development (Delcenserie, 2016). As with studies of language abilities, studies of academic achievements vary considerably in terms of the outcome found (Dalen, 2001; Dalen & Rygvold, 2006; Delcenserie et al., 2013), with a majority of IA children scoring in line with NA peers in terms of language proficiency up to the age of 11 years but some IA children lagging behind (Rygvold & Theie, 2016). Additionally, it has been found that IA children need more special-needs education than their NA peers (Dalen & Theie, 2019; van Ijzendoorn et al., 2005).

In most studies of language development and language performance in IA children, great variety has been found within the group (Scott et al., 2011). Typically, some children performed on a par with their NA peers while some lagged behind or had a language disorder. However, there seems to be a higher risk for IA children to have a delayed language development and language difficulties later in life (Dalen, 2001).

Cleft lip and palate

Cleft lip and palate (CLP) is one of the most common congenital malformations in children, with a global incidence of 1:700 (Mossey et al., 2009). CLP can be associated with other congenital anomalies, but in most cases it is non-syndromic (Mossey et al., 2009). There are different types of CLP: cleft lip, cleft palate, and cleft lip *and* palate. Further, clefts involving the lip and/or the palate can be unilateral or bilateral. Additionally, the extent of an isolated cleft palate can vary from incomplete partial cleft palate to complete cleft palate (Figure 1). The incidence differs between countries and continents; some countries in Latin America and Asia seem to have a higher rate of cleft lip with or without cleft palate than other countries (Mossey et al., 2009). In Sweden, an overall incidence of 2/1000 live births has been reported for CLP (Hagberg et al., 1998). The incidence of different types of cleft also varies; unilateral CLP (UCLP) is the most common one (Hagberg et al., 1998; Mossey et al., 2009). Finally, across ethnic groups, cleft lip with or without cleft palate seems to be more common in males than in females, whereas an isolated cleft palate seems more frequent in females (Hagberg et al., 1998; Mossey et al., 2009).

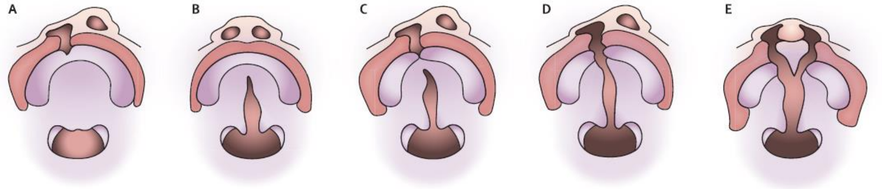

Figure 1. Orofacial clefts: (A) Cleft lip and alveolus. (B) Cleft palate. (C) Incomplete unilateral cleft lip and palate. (D) Complete unilateral cleft lip and palate. (E) Complete bilateral cleft lip and palate. This figure was published in *Orthodontics and Occlusal Management*, Shaw W, Copyright Elsevier (1993). Reprinted with permission.

Clinical practice for children with CLP

Around the world, opportunities for children with CLP to undergo multidisciplinary interventions vary a great deal. In many countries, there are limited opportunities, or none at all, to have palatal surgery closing the cleft lip and/or palate. Early palatal surgery is crucial for optimal speech and language development and necessary to attain functional speech for communication. However, there is as yet no consensus internationally about the optimal age for palatal closure (Lohmander, 2011).

All children with CLP in Sweden receive treatment from a multi-disciplinary CLP team at a tertiary hospital located near the child’s home. Those CLP teams include speech-language pathologists (SLPs), plastic surgeons, orthodontists, nurses, psychologists and audiologists working together to ensure the best possible care for the children and their families. There are six CLP teams in Sweden and they all follow national guidelines for the follow-up of children with CLP (CLP registry, 2020). According to those guidelines, routine assessments/follow-ups are offered to children until the age of 19 years. The assessments are standardised, and the family comes into contact with the CLP team when the child is newborn, if the birth takes place in Sweden. In fact, some families whose child has had a cleft verified through prenatal ultrasound meet with the team even before birth. During the assessments, the child and her/his parents individually meet all professions included in the CLP team for follow-up to ensure the best possible development for each child. The national guidelines include standardised speech assessments performed every two years until the age of 7 and then every three years until the age of 19. The speech assessments follow a specific protocol including audio recording of the Swedish articulation and nasality test (SVANTE; Lohmander et al., 2015). Information about speech and language stimulation is given early on to help parents stimulate their child's speech and language development, and later on speech-therapy intervention is given when needed. However, there are some differences between teams.

The *surgical interventions* performed to close a cleft palate in Sweden differ between hospitals. Two surgical approaches are used. At two treatment centres (Linköping and Malmö), a one-stage procedure to close the soft and hard palate is performed when the child is 9–15 months old (CLP registry, 2020; Lohmander, 2011). At the other four hospitals (Gothenburg, Stockholm, Umeå and Uppsala/Örebro), a two-stage surgical procedure is used instead, with lip, nose and soft-palate repair at the age of 6 months and hard-palate closure at the age of 2 years. The children included in Studies I–III in the present thesis had all undergone a two-stage procedure in accordance with the Gothenburg protocol for surgical interventions (Lilja et al., 1996). Of the children included in Study IV, about three-quarters had done so as well whereas most of the remaining one-quarter had undergone a one-stage procedure.

Hearing is often affected in children with CLP; conductive hearing loss due to middle-ear conditions such as otitis media with effusion (OME) is more common in children with CLP than in non-CLP peers (Purdy et al., 2019). Middle-ear conditions such as OME tend to fluctuate both in frequency and in severity, but OME most commonly causes hearing loss in the mild range (Purdy et al., 2019). However, pre-school children with CLP have an increased risk of developing more persistent hearing problems due to OME than non-cleft children (Flynn et al., 2009), which is why hearing and ear status is regularly assessed and included in the routine follow-up protocol for children with CLP.

Speech and language development in children with CLP

A complete palate and the ability to separate the oral and nasal cavities from each other by closing the passage between them are among the prerequisites for a functional speech ability (Chapman & Willadsen, 2011). Some children with CLP may still have speech difficulties even after primary palatal closure has been performed; there are reports of around 50% of all children with UCLP having speech difficulties at the age of 5 years (Britton et al., 2014; Sell et al., 2017; Willadsen et al., 2017). In most cases, such difficulties are due to a residual cleft in the hard palate, to fistulae and/or to velopharyngeal (VP) incompetence (Chapman & Willadsen, 2011). However, there are differences depending on the child's age and on the time elapsed since palatal repair, with difficulties decreasing by age and by time passed after palatal repair. Additionally, other variables (e.g. cleft type and severity) may also have an impact on speech development (Chapman & Willadsen, 2011).

Although there is a lack of evidence and no clinical consensus internationally regarding the age when early palate closure should be performed to obtain the best possible speech outcome, in theory the palate should be closed as early as possible (Kemp-Fincham et al., 1990). There are a few studies on late palatal repair in children above the age of 8, in adolescents and in adults (Bruneel et al., 2017; Schönmeyr et al., 2015; Sell & Grunwell, 1990) which show only a limited effect on speech outcomes after palatal surgery and high rates of remaining symptoms of VP incompetence.

Speech difficulties in children with CLP are usually articulation errors (placement errors) and errors related to an incompetent VP function (Chapman & Willadsen, 2011). The articulation errors are thought to be related to the cleft palate as such and are therefore often referred to as 'active cleft-speech characteristics' (active CSCs) (Harding & Grunwell, 1998). These errors can be broken down into oral (pertaining to the oral cavity) and non-oral ones. Oral retracted articulation, in which the place of articulation for anterior speech sounds is replaced with a palatal/velar/uvular place of articulation, is a typical example of an oral CSC. Typical non-oral CSCs are glottal and pharyngeal articulation and active nasal fricatives. Active non-oral CSCs are usually due to mislearning or to a strategy intended to compensate for an inadequate VP closure (Hutters & Brondsted, 1987), while active oral CSCs are very common in children with a residual cleft or fistula in the hard palate (Lohmander et al., 2006; Willadsen et al., 2017). Other errors related to an incompetent VP function, which are often referred to as passive articulation errors (Harding & Grunwell, 1998; Hutters & Brondsted, 1987), include hypernasality, audible nasal air leakage and weak pressure consonants.

Most studies of children with CLP have focused on speech development and speech production, even though some studies of the expressive language of toddlers and preschool children with CLP (Hardin-Jones & Chapman, 2014; Scherer & D'Antonio, 1995) have highlighted that a restricted consonant inventory is associated with a risk of delayed expressive development. The phenomenon of lexical selectivity (Vihman, 1993) may have a particular impact on expressive development in children with CLP. Lexical selectivity means that children – regardless of whether they have CLP – tend to choose their very first words based on the speech sounds that they are able to produce or articulate. Lexical selectivity has been studied in children with CLP (Willadsen, 2013) and has been suggested as a possible explanation for delayed expressive language in this group of children.

Language difficulties such as late emergence of one- and two-word phrases and shortness and reduced syntactical complexity of utterances have been reported by several studies (Broen et al., 1998; Hardin-Jones & Chapman, 2011; Kuehn & Moller, 2000; Lamônica et al., 2016; Scherer & D'Antonio, 1995). One Swedish study reported a high prevalence of phonological simplification processes in 5-year-old children with UCLP (Klintö et al., 2016), although other reports concerning children above the age of 3 years have shown ambiguous results regarding language ability (Cavalheiro et al., 2019; Chapman, 2011; Collett et al., 2010a; Klintö et al., 2015; Konst et al., 2003). Some studies of children with CLP aged 4–6 years have reported significantly lower expressive and receptive language ability compared with age- and gender-matched peers without CLP (Cavalheiro et al., 2019) as well as a high frequency of language difficulties in tasks of expressive grammar and vocabulary among children with CLP (Young et al., 2010), although other studies have reported no significant differences on tasks of expressive language between children with and without CLP (Chapman, 2011; Collett et al., 2010a). Klintö et al (2015), who used an expressive-language measure called the Bus Story Test, found a strong trend for a weaker ability to retell information in 5-year-old children with UCLP compared with children without CLP.

Studies of older children around the age of 7–10 years are more scarce, but those that exist have continued to report ambiguous language outcomes. No significant differences in expressive and receptive language ability were found between children with CLP and a group of children without CLP which was matched for gender, age and maternal level of education (Boyce et al., 2018). Another study reported language skills within average ranges (Saervold et al., 2019). By contrast, other reports have found children with CLP to manifest a high frequency of language impairment (Morgan et al., 2017), significant difficulties in language ability compared with test norms (Ghayoumi Anaraki et al., 2016) and significantly lower expressive language ability (measured using tests of word naming and sentence repetition) compared with non-CLP controls (Conrad et al., 2009).

Variability of findings is also characteristic of studies of various aspects of reading and writing in children with CLP. Some studies report no increased risk of reading disability in children with CLP (Collett et al., 2010b) while others do (Collett et al., 2010b; Conrad, 2019; Conrad et al., 2014).

Speech and language assessment

Speech production

Speech production – that is, articulation proficiency and velopharyngeal competence – can be assessed using many different methods, which offer different levels of detail and are suited for different purposes in speech research. The most common methods are perceptual assessment and phonetic transcription of single words and continuous speech (McLeod & Baker, 2017). While speech production is in fact one of the primary outcome measures in the clinical management of CLP, there remains great variety in the methods used to assess and analyse this variable both in research contexts and in clinical reports, as is abundantly clear from the review of reports of speech outcome in IA children with CLP presented below (see Table 1). The gold standard for perceptual speech assessment in cleft speech research is usually to report the outcomes of independent analyses of phonetic transcriptions or ratings performed by at least two trained and blinded SLP listeners/assessors/raters (Lohmander & Olsson, 2004; Sell, 2005; Wyatt et al., 1996). It is also mandatory to use standardised assessment protocols (Kuehn & Moller, 2000) and to report inter- and intra-rater reliability measurements for ratings and transcriptions (Wyatt et al., 1996). Ratings of articulation errors may also be seen, although not as frequently as in the past (Lohmander & Olsson, 2004); those may offer less detailed analyses of articulation than phonetic transcriptions.

For the study of speech production in the present thesis, it was decided to focus on consonant proficiency, on the type and number of consonant errors, on consonant inventory and on perceived velopharyngeal competence. Measures of proficiency at consonant production are commonly used in cleft speech research today (Sell & Sweeney, 2019; Willadsen et al., 2017). One frequent measure is percent consonants correct (PCC), which was originally developed for the assessment of speech disorders not related to cleft palate and was also developed in order to grade the severity of disorder (Shriberg & Kwiatkowski, 1982). In the original work of Shriberg and Kwiatkowski (1982), PCC was calculated on the basis of children's continuous speech and used as a measure of the severity of phonological disorders. PCC assigns equal weight to all speech errors regardless of their aetiology. In cleft speech research, a modified version of the PCC measure is typically calculated on the basis of the target consonants included in a speech material consisting of single words (Allori et al., 2017): the number of correctly produced target consonants is divided by the total number of target consonant and then multiplied by 100. The International Consortium for Health Outcome Measurements recommends that PCC should be used as an outcome measure for articulation proficiency when reporting speech outcome in children with CLP (Allori et al., 2017).

A number of additional measures have been derived from the PCC (Shriberg et al., 1997). One of them, percent consonant correct adjusted for age (PCC-A), has begun to be used more frequently in recent years to measure consonant proficiency in children below school age (Klintö et al., 2019). In the PCC-A, considerations regarding typical and atypical speech development are made. Errors that are considered typical (and hence not atypical) at a certain age are counted as correct if the child is below or at that age. For example, distortions of the /s/ phoneme are considered to be very common in 3- and 5-year-old children with a typical speech development. For this reason, /s/ errors are 'overlooked' and counted as correct in the calculation of PCC-A at those ages, so as to adjust the PCC to reflect typical speech development. In the present thesis, normative data for the SVANTE (Lohmander et al., 2015) and from Lohmander et al. (2017a) were used to establish rules for calculating PCC-A.

Two other common measures of articulation proficiency derived from the PCC are percent correct place of articulation (PCP) and percent correct manner of articulation (PCM) (Klintö et al., 2011; Lohmander & Persson, 2008; McLeod & Baker, 2017). They are calculated by dividing the number of instances of a correct place or manner of articulation by the total number of targeted consonants and then multiplying by 100. To understand the concepts of place and manner of articulation, note that most consonants are produced when an airstream from the lungs is interfered with in some way. The place of articulation refers to the place in the vocal tract where the articulators (the tongue, the lips, etc.) stop, constrict or slow the airstream. The manner of articulation refers to how the consonant is produced, that is to the type of constriction made in the vocal tract (McLeod & Baker, 2017).

Apart from analysing proficiency, another approach in cleft speech research is to perform an error analysis. There is a range of categorisations available for this purpose, and several of them have been used in the present thesis. In European cleft speech research, there is a tradition of categorising errors related to the cleft palate as different types of cleft speech characteristics (CSCs). CSCs are most often divided into active and passive ones (Hutters & Brondsted, 1987). Active CSCs can be described as compensatory errors that are direct consequences of an inability – due to a residual cleft, a fistula or an incompetent VP function – to build the intra-oral pressure required to produce oral speech sounds (consonants). Active CSCs are broken down into non-oral and oral errors. Typical non-oral errors are glottal and pharyngeal articulation and nasal fricatives, while a common oral error is retracted oral articulation, meaning that consonants normally produced at the front of the oral cavity are instead produced further back. In the present project, retracted oral articulation is defined as cases where anterior consonants are retracted to a palatal, velar or uvular place of articulation. The definitions and the delimitation of categories may vary between studies and countries (Sell, 2005), but most recent studies originating from the Scandcleft trials (Willadsen et al., 2019; Willadsen et al., 2017) have used similar definitions to the ones used here. The other category of CSCs, passive ones, includes errors not deemed to be the result of active efforts to compensate. Types of passive CSCs commonly found in children with CLP are hypernasality, errors affecting consonants with nasal emissions and nasal turbulence (audible nasal air leakage), nasalisation of vowels and voiced consonants, and reduced pressure on obstruent consonants (weak pressure consonants) (Chapman & Willadsen, 2011).

Consonant errors can occur in the typical speech development of preschool children (Lohmander et al., 2017a). For example, the replacement of specific consonants – such as changing the place of articulation from velar [k] to dental [t] – is a typical developmental speech error found in many children around the age of 3 years. This type of consonant error is not thought to be related to any structural anomaly in the oral cavity, but rather to be the result of an immature or developing speech-sound system. In the present thesis, the term developmental speech characteristics (DSCs) is used for such errors; they are defined as consonant errors not believed to be related to the cleft palate. A similar analysis and description of DSCs has been used in recent studies of cleft speech (Willadsen et al., 2019; Willadsen et al., 2017).

Velopharyngeal competence

Velopharyngeal (VP) competence can be analysed instrumentally (for example using nasendoscopy, videofluorscopy or magnetic-resonance imaging) to visualise the VP mechanism (Sell & Pereira, 2011), but this has not been able to replace perceptual assessment by trained listeners as the gold standard in the clinical management of CLP and in research (Sweeny, 2011). Methods involving overall ratings and different rating scales are often used, although there is substantial variation in the level of agreement of listeners' ratings of nasality (Sweeny, 2011). A number of objective factors are known to influence ratings, including the listening conditions (Sell et al., 2009), the raters' training (Lee et al., 2009) and the speech sample used (Sell et al., 2009). Such factors may affect the reliability of ratings and yield low levels of inter- and intra-rater agreement and there are many reports on the difficulties of reaching acceptable agreement for ratings of hypernasality (Brunnegård & Lohmander, 2007; Lohmander et al., 2017b).

Because of the low reliability of ratings of hypernasality, combined approaches to assess VP function have evolved (John et al., 2006). One way of attaining a more reliable and valid assessment is to combine perceptual assessment of hypernasality with calculations based on phonetic transcriptions of speech errors related to an incompetent VP function (i.e. weak pressure consonants, audible nasal air leakage, active non-oral errors). VPC-Sum is one example of such a method which has been widely used (Pereira et al., 2013) and found to have satisfactory validity (Lohmander et al., 2917c); it has also been recommended for use in research (Lohmander et al., 2017c) and is used in the present thesis.

Language ability

Different aspects of language ability can be assessed using different methods and tests, but it is a common standard both clinically and in research to measure receptive and expressive language using standardised tests. There is still a lack of normed Swedish-language tests of children's language abilities, even though a few standardised tests are available. One of them is the Test for Reception Of Grammar, TROG-2, which has Swedish normative values (Bishop, 2009). A measure of receptive grammar is necessary in order to identify a potential language disorder and a crucial measure in both clinical and research-related language assessments (Paul et al., 2018). The Clinical Evaluation of Language Fundamentals (fourth edition) (CELF-4) is a comprehensive standardised test that includes measures of receptive and expressive language (Semel, Wiig, & Secord, 2013). This test is often used in child-language research and has also been used in one previous study of IA children with CLP (Morgan et al., 2017). Expressive language is a broad term covering many areas of language, but it usually includes morphosyntax and vocabulary. Tasks of sentence repetition (such as the Recalling Sentences subtest of the CELF-4) capture morphosyntactic ability in children but are also considered to reflect their overall language ability (Klem et al., 2015) and could therefore be a suitable measure to identify language disorders (Conti-Ramsden et al., 2001; Vang Christensen, 2019).

When it comes to gathering data, audio- and video-recording are common ways to capture spontaneous language which have been used both by researchers in the field of cleft-palate speech and in studies of internationally adopted children (Gauthier et al., 2012; Scherer et al., 2018), for example as a basis for measuring MLU (Glennen, 2014). MLU is a classic measure of morphosyntactic ability and has also been found to be low in IA children (Glennen, 2014). A measure similar to MLU is sentence length as captured in the Bus Story Test (Renfrew, 2002), which was originally developed as a screening instrument to identify language disorder but has also been used as a measure of narrative retelling with pictorial support. In one study of narrative retelling in NA children with UCLP, Klintö et al. (2015) analysed samples elicited by the Swedish version of the Bus Story Test, and that test has also been shown to predict academic performance (Bishop & Adams, 1990; Stothard et al., 1998).

To increase the validity of language assessments, it has been suggested that parental questionnaires should be used (Bishop & McDonald, 2009). One such questionnaire developed to help clinicians assess and identify a language disorder and to gain insight into the everyday effects of a language disorder is the Children's Communication Checklist (CCC-2), which is a validated 70-item questionnaire with different statements about the speech, language and communicative abilities of children in everyday life (Bishop, 2012). The CCC-2 has also been used in clinical research relating to children with CLP and/or related syndromes (Boyce et al., 2019; Van Den Heuvel et al., 2017).

Speech and language development in internationally adopted children with cleft lip and palate

So far, few studies have been carried out into the speech and language development of IA children born with CLP. What is more, an evident problem with a majority of those few studies is that they are retrospective studies involving medical chart reviews and covering many years of clinical practice (Table 1).
With a retrospective chart review, there is no possibility to ensure that a variety of standard demands made of research are met, for instance when it comes to the use of standardised protocols and assessments, independent blind analyses performed by trained SLPs not involved in treatment, or reliability measurements. Further, it is not clear from many of the existing studies what methods of assessments were used or at what ages assessments were performed (Goldstein et al., 2014; Swanson et al., 2014). In addition, some studies report data on speech outcome although they lack complete data (Shay et al., 2016; Werker et al., 2017). Even so, despite the limited quality of the scientific data presented, which makes it impossible to compare findings across studies and to draw firm conclusions, it should be noted that most prior studies do report that IA children need secondary VP surgery more often than NA children.

A total of four studies (Morgan et al., 2018; Morgan et al., 2017; Sahlsten Schölin et al., 2020; Scherer et al., 2018) have so far reported on speech and/or language development in IA children in detail and using standard methods of assessment and analysis.

Morgan et al. (2017) performed a detailed cross-sectional study on language development in 51 IA children with CLP at early school age, comparing them with same-age NA peers with CLP (mean age approximately 6 years). Most of the children had been adopted from China and their mean age at palatal repair was approximately 2 years. After controlling for factors such as socioeconomic status, gender, age at assessment, non-verbal IQ and hearing status, the authors found that the IA children scored significantly lower on the language measures tested (receptive language, expressive language, language content and language structure) using the Clinical Evaluation of Language Fundamentals (CELF-P2 and CELF-4), a standardised test. They also found that a younger age at adoption was associated with better language performance. However, the frequency of language disorder was high among both IA (31%) and NA (20%) children.

Scherer et al. (2018) performed a longitudinal pilot study of four IA children with CLP (aged 19–38 months), who were assessed at three different timepoints during a period of 10–12 months, and found that the adopted children had significantly lower scores on speech and expressive language measures than NA peers matched for age, gender and cleft type. This was generally true for all timepoints. The IA children had lower PCCs and smaller consonant inventories at all timepoints. A smaller vocabulary and a lower MLU were also found at timepoint 3, around a year after the first assessment. The authors did not find any differences between the groups with respect to cognitive skills or receptive-language measures, but measures of speech and expressive language showed that the gap between IA and NA children seemed to increase by age.

Morgan et al. (2018) investigated articulation proficiency in 51 IA children with CLP compared with 65 NA children with CLP at the age of 3–9 years. The mean age at assessment was approximately 6 years for both groups. The authors also studied the impact of international adoption on age at primary palatal surgery and on VP competence. The IA children had poorer articulation skills than the NA children and there was a tendency for the IA children to produce more cleft-related speech errors than the NA children. Further, the authors suggested that the low articulation scores of the IA children were related to a higher rate of velopharyngeal incompetence (or 'velopharyngeal insufficiency', VPI) and that 'ongoing VPI has a detrimental effect on articulation skills' (Morgan et al., 2018, p. 8). An additional finding was that the IA children required secondary VP surgery more often than the NA children.

On a related note, Sahlsten Schölin et al. (2020), a recent Swedish study, also found a higher frequency of VP incompetence in IA children than in their NA peers. Comparing IA children with CLP with a group of NA children matched for age, gender and cleft type with respect to ratings of articulation errors and overall VP function, the authors also found no significant differences between the IA and NA children on the two articulation errors that were rated (glottal articulation and oral retracted articulation), which was also in line with the findings of Morgan et al. (2018).

Table 1. Review of studies of articulation, velopharyngeal competence and language in internationally adopted children with cleft lip and palate

Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Goldstein et al. (2014)	Retrospective review of medical records	109 IA children with cleft lip with or without cleft palate = All patients at a US clinic between May 1993 and August 2010 *Origin*: China most common country of origin (78%) *Cleft type*: UCLP most common type *Age at adoption*: 30.5 months *Age at speech assessment:*	Outcomes were compared between groups (pre-adoption and post-adoption surgical outcomes) Demographic data Descriptive statistics	***Articulation***: - ***VPC***: Hypernasality: 41% underwent pre-adoption palate repair, 61% underwent post-adoption palate repair. VPI documented using nasendoscopy: observed in 14% (n=3) with pre-adoption palate repair vs. 19% (n=7) with post-adoption palate repair. Secondary surgery for VPI: 15% ***Language***: -
Sullivan et al., (2014)	Retrospective review of medical records	55 IA children with CLP who underwent two-flap palatoplasty in 1984–2012 at a US clinic *Origin*: China most common country of origin (65%) *Cleft type*: UCLP most common type *Age at adoption*: - *Age at speech assessment:* 4 years	Outcome measures: - perceptual speech results - complications Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence Demographic data Descriptive statistics	***Articulation***: Palatal repair was usually performed at an advanced age and often resulted in a poor speech outcome. ***VPC***: 49% = borderline to borderline incompetent to incompetent VP function. Significant association between older age at the time of palatoplasty and risk of VPI Secondary surgery for VPI: 49% ***Language***: -

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Swanson et al. (2014)	Retrospective review of medical records	216 IA children with CLP = all such children at a US clinic in 1997–2011 *Origin*: China most common country of origin (80%) *Cleft type*: UCLP most common cleft type *Age at adoption*: median 2.0 years *Age at speech assessment:*	Demographic data Descriptive statistics	***Articulation***: 7% articulation errors at initial speech and language assessment ***VPC***: VP dysfunction in 48% at first visit to US clinic of those with pre-adoption palatoplasty and in 36% of those having undergone post-adoption palatoplasty at another US clinic. Secondary surgery for VPI: 49% ***Language***: 16% language-acquisition delay at initial speech and language assessment.
Follmar et al. (2015)	Retrospective review of medical records	All patients (n=201) who underwent primary cleft-palate repair at a US clinic in 1993–2006. Of those, 16 were IA and another 2 were NA but had had delayed repair and were included in the delayed-repair group. *Origin*: China most common country of origin. *Cleft type*: UCLP most common cleft type *Age at speech assessment:* 'At least 5 years of speech' at follow up. Mean follow-up 9.3 years.	Primary outcome = VPI Standard-repair group vs. delayed-repair group. Demographic data Descriptive statistics	***Articulation***: - ***VPC***: VPI in 13% (23 of 183 patients) in the standard-repair group vs. 33% (6 of 18 patients) in the delayed-repair group. Secondary surgery for VPI: Standard-repair group 10%, delayed-repair group 6%. ***Language***: -

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

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Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Shay et al. (2016)	Retrospective review of medical records	All IA children (n=151) with CLP at a US clinic in 1998–2012 *Origin*: China most common country of origin (81%) *Cleft type*: UCLP most common cleft type *Age at adoption*: 2.3 years *Age at assessment:* Two years after primary palatoplasty (scores only for 32% of the children), mean age 3.46 years. After the age of 5 years (scores only for 52%), mean age 8.98 years. Complete scores for both periods for only 22%.	Long-term speech outcomes, reported within two years after primary palatoplasty and after the age of 5 years. Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence	***Articulation***: - ***VPC***: Within 2 years after primary repair, 46.7% had competent to borderline competent VP mechanisms After the age of 5 years, 56.8% had competent to borderline competent VP mechanisms Secondary surgery for VPI: 14.8% (n=21) ***Language***: -
Werker et al. (2017)	Retrospective review of medical records	136 IA children = all IA children with cleft lip and/or palate at a Dutch clinic in January 1994–December 2014 *Origin*: China most common country of origin (89%) *Cleft type*: UCLP most common cleft type *Age at adoption*: 26.5 months *Age at assessment:*	Nasal resonance assessed using nasometry Understandability levels scored both by parents and by SLPs. Limited data. Out of 136 children, 13 children were assessed using nasometry before and after pharyngoplasty. Incomplete data regarding understandability as well. There were data on 26 and 35 out of 136 children before and after pharyngoplasty, respectively.	***Articulation***: - ***VPC***: Scores on both nasal and denasal texts improved in all children after pharyngoplasty. Speech improvement (understandability) in 73% and 77% of the patients as assessed by the parents and by SLPs, respectively. ***Language***: -

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Morgan et al. (2017)	Prospective cross-sectional comparison study	Children with CLP were recruited from a paediatric hospital as part of a larger longitudinal study of children born with CLP. *Origin*: China most common country of origin *Age at adoption*: 25.71 months *Ages at assessment:* 3;0–9;0	Comparison of language development in children with CLP with early exposure to English (EE, n=67) vs. late exposure to English (LE, n=51) Mean language scores (CELF-P2, CELF-4) Frequency of language disorder	***Articulation***: - ***VPC***: - ***Language***: LE children scored less well on all language indices. 53% of the EE children and 57% of the LE children obtained standard scores above 90 on all indices. Language disorder: LE: 31%, EE: 20% Younger age at adoption associated with better language skills.
Morgan et al. (2018)	Prospective cross-sectional comparison study	Children with CLP between the ages of 3 and 9 years 51 IA and 65 NA children between the ages of 3 and 9 years with non-syndromic cleft palate with or without cleft lip. *Origin*: China most common country of origin *Age at adoption*: 2.14 years *Age at assessment:* IA: mean 6.54 (3.29–8.99), NA: mean 5.95 (3.37–8.99)	Articulation development VP function Persistent VP dysfunction Goldman–Fristoe Test of Articulation, 2nd Edition (GFTA-2) standard score and cleft-related articulation errors (CREs). VP gap size during closure attempts – VP sufficient or VP insufficient	***Articulation***: Articulation impairment: NA children: 40–76% IA children: 71–92% (depending on age). IA children were not significantly more likely to make CREs. ***VPC***: Age at the time of assessment and VPS were associated with CREs. Higher frequency of need for VPI surgery among IA children (46% vs. 28%) ***Language***: -

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Scherer et al. (2018)	Prospective longitudinal descriptive study	Four IA children with CLP (age range 19 to 38 months) and four NA children Matched for age, gender and cleft type *Origin*: China *Age at adoption*: 9–15 months *Age at assessment:* Assessed at three timepoints over a period of 10 to 12 months.	Risk of delayed speech and language development Speech and language ability analysed using standardised tests, parent surveys, language samples and single-word phonological assessments PEEPS, PLS-4, MCDI PCC, consonant inventory, percent compensatory errors, MLU, intelligibility	***Articulation***: IA: lower number of consonants, limited variety of consonants, lower PCC at all timepoints ***VPC***: Nasal substitutions prevalent in three-quarters of IA children at last timepoint. ***Language***: The mean scores of the IA children were lower than those of the NA group at all three time points for expressive language and speech-sound production. No differences observed in cognitive performance and receptive language measures.
Pet et al. (2018)	Retrospective review of medical records	All patients born between January 2004 and August 2012 who underwent primary repair of CLP at a US clinic, n=84 IA children Speech outcomes: comparison group consisted of 45 IA and 111 NA children *Origin*: Asian most common *Cleft type*: UCLP most common cleft type *Age at assessment:* At or near (within 2 years of) the age of 5 years	VPI, compensatory articulations, nasal air emission and speech intelligibility. Rated on a five-point scale.	***Articulation***: Compensatory articulations, intelligibility and nasal air emission: no significant difference between IA and NA. ***VPC***: VPI (moderate to severe): IA children 16%, NA children 6%. Nasal air emission, no significant difference. Secondary VP surgery: IA 17%, NA 12%, no significant difference ***Language*: -**

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

Article	*Study design*	*Study population*	*Outcome measures*	*Results*
Kaye et al., (2019)	Retrospective review of medical records	All IA patients with CLP at a US clinic in 2004–2014 (n=122) *Origin*: China most common country of origin *Cleft type*: UCLP most common cleft type *Age at adoption*: 26 months *Age at assessment*: Patients presenting for initial or follow-up assessment	Primary measures: number of IA children with CLP in 2004–2014. Secondary measures: number of children with different cleft types, children with other diagnoses, previous cleft-related surgery, post-adoption cleft-related surgery and speech outcomes.	***Articulation***: Articulation disorders: 79.2% (mostly moderate to severe). ***VPC***: VPI 36.6% (43.2% borderline or mild, 45.9% moderate to severe). Secondary VP surgery: 51.3% ***Language***: Speech and language delays: 24.8%, expressive delays: 92%, moderate to severe receptive delays: 80%
Sahlsten Schölin et al. (2020)	Cross-sectional comparison study	n=25 IA children with CLP, born in 1994–2005 Matched for age, gender and cleft type with 25 Swedish-born children. *Origin*: China most common country of origin *Cleft type*: UCLP most common cleft type *Age at adoption*: - *Age at assessment*: 5 years	Perceptual analysis of audio recordings performed by two experienced, blinded SLPs. VPI rated on a three-point scale and articulation on a five-point scale.	***Articulation***: Glottal articulation: no significant difference between groups. Retracted articulation: no significant difference between groups ***VPC***: VPI: rate of occurrence higher among the IA children Secondary surgery for VPI: IA 28%, NA 4% ***Language***: -

Abbreviations: CELF = Clinical Evaluation of Language Fundamentals, CLP = cleft lip and palate, CRE = cleft related errors, EE = early exposure to English, IA = internationally adopted, LE = late exposure to English, MCDI = MacArthur-Bates Communication Development Inventory, MLU = mean length of utterance, NA = non-adopted, PCC = percent consonant correct, PEEPS = Profiles of Early Expressive Phonological Skills, PLS-4 = Preschool-Language Scale–4th Edition, SLP = speech-language pathologist, UCLP = unilateral cleft lip and palate, VP = velopharyngeal function, VPC = velopharyngeal competence, VPI = velopharyngeal insufficiency, VPS = velopharyngeal sufficiency

Rationale for the studies in the present thesis

Previous studies have suggested a high risk of velopharyngeal incompetence in internationally adopted children with cleft lip and palate, although most of those studies unfortunately used an insufficient methodology to investigate velopharyngeal function. The few studies that examined speech production and language in detail found that internationally adopted children may exhibit difficulties with articulation and expressive language up to the age of seven or eight years, but their results are inconsistent. Further, most previous investigations suggested that there is an increased risk of delayed speech and language development in internationally adopted children with cleft lip and palate. Earlier studies of internationally adopted children *without* cleft lip and palate follow the same path, with inconsistent results depending on the methodology used and the age groups investigated but nevertheless a suggestion that there may be a risk of difficulties in language ability in that group as well. To date, there is clearly a knowledge gap; only very few previous investigations were performed in accordance with well-established and preferable standards of methodology.

Velopharyngeal incompetence has a severe effect on a child's articulatory proficiency and intelligibility. Further, a delayed language development may also have a severe impact on a child's learning and future academic performance. An impaired speech production and language ability are also likely to affect a child's social communication and participation in society. Hence there is a need for more studies to perform further detailed investigations of speech-production development in this group of children and to compare their development with that of appropriate groups of peers. Some of the questions that need answers are whether internationally adopted children with cleft lip and palate perform worse in speech production than non-adopted peers with the same cleft type, and whether internationally adopted children with and without cleft lip and palate perform similarly on measures of language ability or whether the children with cleft lip and palate are more severely affected because, besides being internationally adopted, they also have cleft lip and palate.

Aims

The main aim of this thesis was to investigate, from a longitudinal perspective, speech-production development and language ability until school age in internationally adopted children with unilateral cleft lip and palate.

The thesis includes four studies, whose specific aims were the following:

I. To investigate consonant proficiency, consonant errors and velopharyngeal competence at the age of 3 years in internationally adopted children with unilateral cleft lip and palate as compared with children born in Sweden with the same cleft type.

II. To investigate consonant proficiency, consonant errors, consonant inventory and perceived velopharyngeal competence at the age of 5 years in internationally adopted children with unilateral cleft lip and palate as compared with children born in Sweden with the same cleft type.

III. To explore and describe speech-production development between the ages of 3 and 7–8 years and expressive language abilities at the age of 7–8 years in internationally adopted children with unilateral cleft lip and palate, and to investigate the relationship between this development and parent-reported speech-and-language status and age at adoption.

IV. To investigate language abilities at the age of 7–8 years in internationally adopted children with and without unilateral cleft lip and palate and to investigate potential associations between age at adoption and time of exposure to Swedish, on the one hand, and school-age language outcome, on the other.

Methods

The data on which the present thesis is based have been collected using a variety of methods and materials. Data have been collected from reviews of medical charts, from standardised audio recordings made during routine speech assessments, from standardised speech and language assessments using valid and norm-referenced test materials, and from parental questionnaires. When it comes to the audio recordings, phonetic transcriptions and perceptual analyses of different variables have been used to measure outcomes. Assessments of inter- and intra-transcriber/rater agreement have also been performed. Additionally, data from two comparison groups have been collected: non-adopted (NA) children with unilateral cleft lip and palate (UCLP) treated during the same period of time and by the same CLP team, and internationally adopted (IA) children without UCLP adopted around the same time as the children with UCLP.

Participants

A total of 87 children were included in this thesis: 36 IA children with UCLP, 22 NA children with UCLP and 29 IA children without CLP (Table 2). All studies included children born in 2006–2010 with UCLP who had been internationally adopted and were recruited from the Cleft Palate Centre at the Sahlgrenska University Hospital, Gothenburg, Sweden. For Studies I and II, the members of the comparison groups consisting of children born in Sweden in 2006–2010 with UCLP were also recruited from the same Cleft Palate Centre. The inclusion criteria were: born in 2006–2010, no severe hearing impairment, and no known intellectual disability or syndrome. An additional inclusion criterion was set for Studies I and II to ensure a more valid assessment of the children, who were rather young at that point: they had to be able to produce at least 50% of the target words in the speech assessment. As a result of this criterion, eight IA children and three NA children were excluded from Study I and one IA child was excluded from Study II. The longitudinal Study III included 17 IA children with UCLP, all of whom also participated in Study II (see Table 2 for more details).

For Study IV, we also recruited participants from the Cleft Palate Centre at the Skåne University Hospital, Malmö, Sweden, and from national Swedish adoption organisations. A total of 27 IA children born in China with UCLP were included in Study IV;

19 of them were recruited from the Sahlgrenska University Hospital and 6 from the Skåne University Hospital. Two children were recruited through national adoption organisations. The comparison group of IA children without CLP used in Study IV consisted entirely of children recruited through three national adoption organisations: Adoptionscentrum, Barnens Vänner and Barnen Framför Allt. Those organisations published information about the project in their member magazines, on their official web sites and using social media. The inclusion criteria for the IA children without CLP were: born in 2007–2011 and having lived in a Swedish-speaking adoptive family for at least two years. The exclusion criteria were: CLP, severe hearing impairment and known syndrome and/or intellectual disability. A total of 29 IA children without CLP were included in the comparison group in Study IV. They originated from different countries and were born in 2007–2011 in Asia (72%), Africa (21%) or Eastern Europe (7%). At a global level, according to many recent studies, most IA children with CLP originate from China (Hansson et al., 2012). For this reason, it was decided to include only children born in China in the study groups. Further, the original aim at the beginning of the project was to include only children without CLP adopted from China in the comparison group for Study IV. However, during the period in question, there were few children adopted from China without CLP, meaning that it was not possible to recruit such a comparison group. It was therefore decided to include children adopted from any country in the comparison group. Figure 2 shows the proportions of international adoptions to Sweden from different continents during the period when most participants in this thesis were adopted (2006–2011).

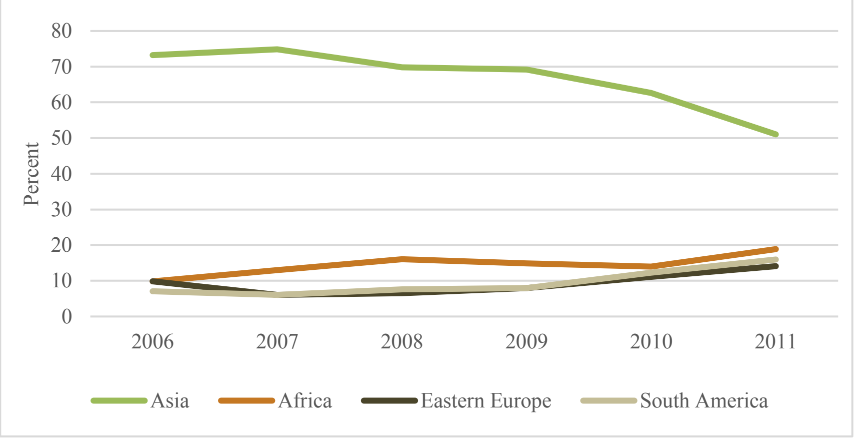

Figure 2. Proportion of international adoptions to Sweden from different continents in 2006–2011. (Family Law and Parental Support Authority, MFoF, 2020)

Table 2. Overview of participants in each study

	Study I	*Study II*	*Study III*	*Study IV*
Study design	Cross-sectional with comparison group	Cross-sectional with comparison group	Longitudinal	Cross-sectional with comparison group
Children's age (years)	3	5	3, 5, 7–8	7–8
Number of eligible IA children	29	31	32	32 (CLP team SahlU) 17 (CLP team SkåU)
Number of IA children who agreed to participate	25	27	17	19 (CLP team SahlU) 6 (CLP team SkåU) 2 (adoption organisations)
Number of IA children included	14	25 (of which 13 children had participated in Study I)	17 (of which 9 had participated in Study I; all 17 had participated in Study II)	27 (of which 17 had participated in Studies II and III, and 9 had participated in Study I)
Number of children in comparison group (children with UCLP born in Sweden)	18 included out of 21 who agreed to participate	20 included out of 20 who agreed to participate (of whom 16 had participated in Study I)		
Number of children in comparison group (IA children without UCLP)				29
Exclusion criteria	Severe hearing impairment (n=0), intellectual disability (n=0), known syndrome (n=0), production of < 50% of the speech material used in speech assessment (n=11 [8 IA, 3 NA])	Severe hearing impairment (n=0), intellectual disability (n=0), known syndrome (n=0), production of < 50% of the speech material used in speech assessment (n=1 [1 IA, 0 NA])	Severe hearing impairment (n=0), intellectual disability (n=0), known syndrome (n=0)	Severe hearing impairment (n=0), intellectual disability (n=0), known syndrome (n=0)

Abbreviations: CLP = cleft lip and palate, UCLP = unilateral cleft lip and palate, IA = internationally adopted, SkåU = Skåne University Hospital, SahlU = Sahlgrenska University Hospital.

Surgical treatment

The children with UCLP included in Studies I–III underwent surgical procedures in accordance with the protocol followed at the CLP centre at the Sahlgrenska University Hospital. The two-stage Gothenburg protocol (Lilja et al., 1996) was performed on all children. It includes lip, nose and soft-palate repair at 6 months of age and hard-palate repair at the age of 2 years. The NA children in this project had their first visit to the CLP centre within approximately three weeks of being born. For the IA children, the first visit to the CLP centre took place approximately three weeks after their adoption. Some children had undergone surgical repair of the lip or both the lip and the palate in China prior to their adoption (Table 3). The IA children who needed surgical repair in Sweden underwent their first procedure after having had two or three months to adjust to their new homes. The repair protocol was not changed for the IA children, but the usual period of four to six months between soft-palate and hard-palate repair, which is intended to facilitate healing, was implemented for them as well. Secondary velopharyngeal (SVP) surgery was performed as soft-palate re-repair using intravelar veloplasty or palatopharyngeal flap.

In Study IV, six IA children with UCLP who had been treated at the Skåne University Hospital also participated. They had been surgically treated (if needed) with palatal closure in a single stage using intravelar veloplasty in accordance with Sommerlad (2003). In addition, two IA children with UCLP who had not been treated either at the Sahlgrenska University Hospital or at the Skåne University Hospital were included. They were recruited through adoption organisations and no data were collected from their CLP teams. However, information about the timing and procedures of surgery was collected from their parents (Table 3).

Hearing status

Information about hearing was collected in two ways in this project (Table 4). For Studies I–III, data from medical charts were accessed; in addition, for most children, an audiometry screening with results in pure-tone average (PTA) had been performed at the hospital close in time to the speech assessments, in accordance with the routine guidelines. In Study IV, information about hearing was collected by means of a parental questionnaire in which the parent(s) had to answer two related items: 'My child has normal hearing (yes/no/I do not know)' and 'My child needs a hearing aid (yes, since the age of X/no)'.

Table 3. Types of surgical interventions and age (in months) at surgery

Study	*Group*	*N*	*Surgical repair in China (lip only/lip and palate), number*	*Age at SPC, mean (range)* [a]	*Age at HPC, mean (range)* [a]	*Age at PC, mean (range)* [b]	*Number of children with SVP prior to speech assessment*	*Number of children with alveolar-cleft repair prior to speech assessment*
I	IA	14	7/4	24 (18–42)	36 (28–55)	-	-	-
	NA	18	-	6 (4–12)	25 (15–37)	-	-	-
II	IA	25	13/5	26 (14–42)	35 (23–55)	-	4	-
	NA	20	-	6 (4–12)	25 (15–37)	-	2	-
III	IA	17	7/3	26 (19–42)	37 (27–55)	-	T1: - T2: 4 (two needed additional surgery between T2 and T3) T3: 2 (one needed additional surgery twice)	T1: - T2: - T3: 3
IV	IA + UCLP	27	14/5	26 (19–42)	37 (27–55)	20 (12–28)	-	-
	IA Non-UCLP	29	-	-	-	-	-	-

Note: [a] For children treated in accordance with the Gothenburg two-step procedure for surgical palatal closure, [b] For children treated in accordance with the one-step procedure for palatal closure.
Abbreviations: IA = internationally adopted, NA = non-adopted, UCLP = unilateral cleft lip and palate, SPC = soft-palate closure, HPC = hard-palate closure, SVP = secondary velopharyngeal surgery, T1 = timepoint 1, T2 = timepoint 2, T3 = timepoint 3.

Table 4. Age (in months) at adoption and hearing status as reported for participants in Studies I-IV

Study	*Group*	*n*	*Gender (♀ / ♂)*	*Age at adoption, mean (range)*	*Hearing, median PTA (L/R), dB*	*Normal hearing (yes/no/do not know) **
I	IA	14	3/11	20 (12–33)	21/22	-
	NA	18	5/13	-	20/20	-
II	IA	25	5/20	21 (7–42)	20/20	-
	NA	20	7/13	-	20/17.5	-
III	IA	17	4/13	22 (16–33)	21/22	-
					20/20	-
					20/20	-
IV	IA + UCLP	27	9/18	22 (10–73)	-	22/5/0[a]
	IA Non-UCLP	29	10/19	22 (7–55)	-	27/1/1[a]

Note: * Hearing status as rated by parents, [a] No child needed a hearing aid.
Abbreviations: IA = internationally adopted, NA = non-adopted, PTA = pure-tone average, L/R = left/right, dB = decibel.

Speech interventions

Data on speech interventions were collected for Studies II and III. The total number of visits to a SLP, including routine visits as well as assessment and speech-therapy/speech-intervention sessions, varied greatly between children. Before the speech assessment performed at the age of 5 years (Study II), the IA children and the NA children had received a similar number of sessions: the median was 9 (min–max 2–26) and 9.5 (min–max 5–23), respectively. As regards the total number of visits between the age of 3 (T1) and the speech and language assessment carried out at the age of 7–8 years (T3) in Study III, the median for the IA children was 15 (min–max 3–37).

Non-verbal cognitive ability

A measure of non-verbal cognitive ability, the Matrix Reasoning subtest from the Wechsler Abbreviated Scale of Intelligence (WASI), was included in Study IV as a background variable (Wechsler, 1999). That subtest yields a raw score, which is converted into a t-score (mean = 50, SD = 10). Since there are no Swedish norms for the WASI, the US norms were used. The reliability coefficients for this subtest range from .86 to .96 for the age groups in question. The mean t-scores of the IA children with and without UCLP were 52 (SD 9.2) and 55.2 (SD 8.5), respectively ($z = -1.42$, $p = .15$).

Ethical considerations

There are specific ethical issues to consider in this project. Any research that includes children and people with somewhat limited autonomy needs to be performed with particular care in terms of the information given, recruitment, inclusion and assessments.

During the period when data were collected and participants were recruited, this project was very positively welcomed by parents of IA children, professionals and adoption organisations. Even so, there was always a risk that the project could be perceived as ‘singling out’ IA children as a group in a negative way. However, it was also evident that there was a lack of knowledge and that the contribution to research could be essential.

Parents were given written information about the project prior to their children’s participation and they were also offered an opportunity to receive oral information. All parents who were legal guardians of potential participants gave their written informed consent for their child to be included as a participant in the project. However, this did not automatically imply that the children themselves would be willing to participate. In Studies III and IV, the author met all families herself and performed all speech and language assessments at the age of 7–8 years. To facilitate participation and to exert as little impact as possible on the children’s everyday life, each family was offered the opportunity to have the assessments performed in a place and at a time that suited them. In most cases, the assessments were performed in the children’s homes at the weekend or on a holiday. The children and their parents always decided together whether the parents would accompany the child during the assessment. In most cases, the parents sat beside their child.

The encounters with the children were carefully designed and implemented. Oral information about the project and tests was simplified to a level adjusted to suit each child and given before the assessments were performed. At each assessment, the children were asked whether they knew anything about the project and about what we were going to do together, and whether they had any questions about the project. This information was also given to the parents before the meeting with the family. During the speech and language assessment, many different tests were used, meaning that each meeting or assessment lasted for approximately 2.5 hours. Pictorial support was used to facilitate the provision of information to the children about what tests, and how many tests, we were going to perform or had already performed, in order for them to gain more knowledge about the procedure. In all cases, a pause of about 15 minutes was included halfway through the assessment meeting, at which time the children were offered a snack. Additional breaks from testing were taken as needed. After the assessment, the children were given a further opportunity to ask questions about the project and to listen to their audio recordings. Finally, each child was given a small toy afterwards.

Besides that toy, the families did not benefit directly from their participation in the project. No financial compensation was offered to those who came to the hospital on a weekday for an assessment. However, the knowledge and evidence gathered through the project will hopefully have an impact on the clinical care provided by CLP teams at hospitals and by SLPs who see children with CLP, including on the procedures used by them, which will benefit other parents in the same situation. Further, the new knowledge will also help ensure that the information given to prospective adoptive parents in the future about speech outcomes and the prognosis for speech development will be more accurate.

Participation in the project entailed little by way of risk for the families and the children. In Studies I, II and III, audio recordings that had been collected previously at the ages of 3 and 5 years during routine visits to the hospital were used. Audio recordings were also used in Study IV. All recordings were coded and randomised, and no personal information was kept in the audio recordings or test protocols. This was mainly done for scientific purposes, but also to ensure privacy.

The studies included in the present thesis were approved by the Regional Ethical Review Board of Gothenburg, Sweden (case numbers 865-13 and T022-16).

Procedure

Data collection

In Studies I–III, material from standardised audio recordings from the routine visits at the ages of 3 and 5 years to the CLP team at the Sahlgrenska University Hospital were used. This material included standardised speech assessments using SVANTE, the Swedish Articulation and Nasality Test (Lohmander et al., 2015), which were performed and recorded by any of the SLPs on the team. Spontaneous speech was also recorded routinely at the ages of 3 and 5 years, but to a varying extent. For this reason, those recordings were used only for perceptual assessments of VP competence in Studies I and III (at T1). The speech and language data for T3 in Study III were in all cases collected by the author. For Study IV, all speech and language data were collected by the author in a location chosen by the parents (such as in the family home or at the hospital). All audio recordings were performed using a portable digital stereo audio recorder (Tascam Hdp2, Teac Corporation) and a microphone (Ecm-ms 957, Sony Corporation).

For Studies I and II, the speech recordings of the speech assessments were edited so that each of the 59 words of the SVANTE (each word including one target sound) constituted a separate .wav file. Hence there was a maximum of 59 .wav files from each child to be phonetically transcribed on each occasion. The editing of the speech recordings was performed using the Praat software (Boersma, 2001) on a PC. Prior to phonetic transcription, each child and his/her .wav files were coded and randomised so that the transcriptions would be performed blindly, without the transcriber knowing whether a given recording was of a child who had been internationally adopted or not. All editing was performed by the author, who was also assigned the role of master transcriber and performed the phonetic transcriptions of all speech material.

In Study III, the speech recordings made at T3 were edited in a similar manner but were not coded or randomised for phonetic transcription, since this was a longitudinal study without a comparison group. However, for the rating of VP function, all recordings were coded and randomised.

In Study IV, the SVANTE recordings were edited by two final-year SLP students, who also randomised the coded .wav files prior to phonetic transcription in order to make the transcription process as blind as possible.

Table 5. Speech and language material, assessment methods, outcome measures and data level for each study

Study	*Age*	*Material*	*Assessment method*	*Outcome measures*	*Data level*
I	3 years	SVANTE words	Phonetic transcription	PCC-A, PCP, PCM	Continuous
				Proportion of children with consistent consonant errors (%)	Continuous
				Consonant errors related to place of articulation: oral retracted articulation, non-oral articulation Consonant errors related to manner of articulation	
				Deletion of target consonants Errors related to voicing	
				Prevalence of audible nasal air leakage	
		Spontaneous speech	Scale rating (yes/no)	Overall VP competence	Binary outcome
II	5 years	SVANTE words	Phonetic transcription	PCC, PCC-A, PCM, PCP	Continuous
				Consonant inventory Number of consonants	Continuous
				Consonant errors: CSCs, DSCs, deletion of target consonants Proportion of children with consistent consonant errors (%)	Continuous
		SVANTE words	Phonetic transcription Scale rating	VPC-Sum 0–6 Proportion of children with competent, marginally incompetent or incompetent VP function (%)	Ordinal scale

Study	*Age*	*Material*	*Assessment method*	*Outcome measures*	*Data level*
III	Pre-adoption	Parental questionnaire		No speech at all Babbling Using words in native language Using sentences in native language Do not know Proportion of children (%)	Ordinal scale
	T1 = 3 years	SVANTE words	Phonetic transcription	PCC	Continuous
				Consonant inventory Number of consonants	Continuous
				Consonant errors: non-oral articulation, retracted oral articulation, deletion of target consonants, developmental speech characteristics Proportion of children with consistent consonant errors (%)	Continuous
		Spontaneous speech	Scale rating	VPC-Rate 0–2 Proportion of children with competent, marginally incompetent or incompetent VP function (%)	Ordinal scale
	T2 = 5 years	SVANTE words	Phonetic transcription	PCC	Continuous
				Consonant inventory Number of consonants	Continuous
				Consonant errors: non-oral articulation, retracted oral articulation, deletion of target consonants, developmental speech characteristics Proportion of children with consistent consonant errors (%)	Continuous
		SVANTE sentences	Scale rating	VPC-Rate 0–2 Proportion of children with competent, marginally incompetent or incompetent VP function (%)	Ordinal scale
	T3 = 7–8 years	SVANTE words	Phonetic transcription	PCC	Continuous
				Consonant inventory Number of consonants	Continuous
				Consonant errors: non-oral articulation, retracted oral articulation, deletion of target consonants, developmental speech characteristics Proportion of children with consistent consonant errors (%)	Continuous
		SVANTE sentences	Scale rating	VPC-Rate 0–2 Proportion of children with competent, marginally incompetent or incompetent VP function (%)	Ordinal scale
		Recalling Sentences (CELF-4 subtest)		Measure of expressive language: Recalling Sentences, scale score	Continuous
		Bus Story Test		Measures of expressive language: Sentence Length, raw score Information, raw score Subordinate Clauses, raw score	Continuous

Study	*Age*	*Material*	*Assessment method*	*Outcome measures*	*Data level*
IV	7–8 years	TROG-2, receptive grammar		Measure of receptive language: TROG-2, Standard score	Continuous
		Recalling Sentences (CELF-4 subtest)		Measure of expressive language: Recalling Sentences, scale score	Continuous
		Bus Story Test		Measures of expressive language: Sentence Length, raw score Information, raw score Subordinate Clauses, raw score	Continuous
		SVANTE words	Phonetic transcription	PCC	Continuous
		CCC-2, subscales		Speech, scale score Syntax, scale score Semantics, scale score Coherence, scale score Initiation, scale score Stereotyped language, scale score Use of context, scale score Non-verbal communicative ability, scale score	Continuous

PCC-A = percent consonants correct, adjusted for age, PCP = percent correct place of articulation, PCM = percent correct manner of articulation, PCC = percent consonants correct, VP = velopharyngeal, VPC-Sum = velopharyngeal composite-score sum, VPC-Rate = velopharyngeal competence rate, CELF-4 = Clinical Evaluation of Language Fundamentals – version 4, BST = Bus Story Test, SVANTE = Swedish Articulation and Nasality Test, TROG-2 = Test for Reception of Grammar – version 2, CCC-2 = Children's Communication Checklist – second version.

Speech material and analysis

Speech production – i.e. consonant proficiency, consonant-error analysis, consonant inventory and velopharyngeal (VP) function – was analysed in Studies I–IV on the basis of phonetic transcriptions of material from the SVANTE and, to some extent, of spontaneous speech. The analysis mainly targeted consonant proficiency and consonant errors. VP function was analysed using different methods in each of Studies I–III.

The SVANTE is a single-word test using picture naming. It includes a total of 59 words to be elicited, each word containing one target consonant. The target consonants represent six different plosives /p b t d k g/ and two different fricatives /f s/. Each consonant is represented three times in word-initial position, twice in medial position and twice in word-final position. The voiceless alveolo-palatal fricative /ɕ/ is represented three times in word-initial position. The SVANTE also includes additional words for assessing nasals, meaning that the full Swedish consonant inventory of 18 consonants can be assessed. Additionally, there are 13 sentences included in the SVANTE where high- and low-pressure consonants and nasal plosives occur together.

Semi-narrow phonetic transcription of the target consonants in each SVANTE word was performed using the IPA alphabet (IPA, 2018) and Ext IPA (ICPLA, 2015). Only the following diacritics were used: [͋] for nasal emission, [͌] for velopharyngeal friction and [͈] for weak articulation; in addition, the symbol [n̥̊] for the non-oral active nasal fricative was used. The author was the master transcriber for all phonetic transcriptions in Studies I–III and transcribed all speech material. In Study IV, two final-year SLP students transcribed all speech material independently. Across all studies, 30% of all material was re-transcribed for the assessment of agreement.

Consonant proficiency and consonant errors

In order to analyse articulation proficiency, different measures of consonant proficiency and consonant errors were used as outcome variables in this thesis (Table 5). Percent consonants correct (PCC) and/or percent consonants correct adjusted for age (PCC-A) was used in all four studies. In Studies I and II, normative data from the SVANTE were used as a guideline when adjusting the PCC measure into PCC-A. Audible nasal air leakage or weak articulation, which can be symptoms of an incompetent VP function, did not affect PCC-A scores. In addition, two other measures derived from the PCC – percent correct place of articulation (PCP) and percent correct manner of articulation (PCM) – were used as outcome variables in Studies I and II.

The types and frequency of consistent *consonant errors* related to place and manner of articulation were analysed in Study I. A consonant error was considered consistent when it occurred on three or more occasions among the target-consonant instances. *Consonant errors related to place of articulation* were described in terms of either of two cleft-speech characteristics (CSCs): non-oral articulation or oral retracted articulation. Non-oral articulation included glottal stops or fricatives and active nasal fricatives. Oral retracted articulation was defined as all errors involving a shift from an anterior to a posterior (palatal to uvular) place of articulation. *Consonant errors related to manner of articulation* included changes from a target plosive consonant to a nasal/fricative/approximant consonant and changes from a fricative target consonant to a plosive consonant. Further, two additional types of consonant error were included in the analyses performed in Study I: deletion of target consonants and errors related to voicing. The frequencies of all consistent consonant errors were calculated and presented.

In Study II, the frequency of consistent consonant errors was also analysed, defined in the same way as in Study I but with a different categorisation of errors. Here, consonant errors were categorised as *CSCs*, *developmental speech characteristics (DSCs)* or *deletion of target consonant*. CSCs were broken down into non-oral articulation (glottal/pharyngeal consonants, glottal reinforcements and active nasal fricatives) and oral retracted articulation (same definitions as in Study I). The DSCs that occurred in the material were velar fronting, stopping and voicing. Deletions of target consonants were also analysed and reported separately.

In Study III, the frequency of four categories of consistent consonant errors was analysed: *CSCs* (glottal/pharyngeal stops/fricatives, glottal reinforcements, active nasal fricatives and target consonants replaced by a nasal consonant), *oral retracted articulation* (same definition as in Studies I and II), *deletion of target consonant* and *DSCs.* The DSCs that occurred in this speech material were the same as in Study II plus approximation.

Consonant inventory was analysed on the basis of the phonetic transcriptions in Studies II and III by calculating the number of established consonants. A consonant was defined as established when its realisations were correct in at least 50% of the possible instances, regardless of position. For some children, not all 18 Swedish consonants were elicited during the SVANTE speech assessment. The consonant inventory was reported in terms of mean and median values along with the percentage of children who had established each consonant.

Velopharyngeal competence

To study VP competence, different types of speech material and different methods were used (Table 5). In Study I, overall perceived VP competence was rated on a two-point scale (competent – not competent) based on spontaneous speech. Separate ratings were performed by three experienced SLPs who were blind as to whether a child was IA or not. To be included in the analysis of VP competence, the child had to produce at least 20 seconds of combined utterances. An utterance was defined as a minimum of a two-word phrase (noun + verb) or the child counting. The assessment material consisted of .wav files each containing all available utterances from a given child which had been combined using the Praat software. Only 11 IA children and 17 children in the comparison group produced enough speech material to be included in the perceptual analysis of VP competence.

In Study II, VP competence was analysed using VPC-Sum (Lohmander et al., 2017c). This method combines a perceptual analysis of hypernasality with calculations of the number of non-oral articulation errors and the number of perceptual symptoms of VP incompetence based on phonetic transcriptions (Table 6). For the perceptual analysis of hypernasality, the first nine words of the SVANTE were combined into a string (a .wav file) using the Praat software. The .wav file for each child was then perceptually analysed by three judges. The nine words were chosen because they include high vowels thought to be vulnerable to hypernasality (Hutters & Henningsson, 2004;

Lohmander et al., 2017b). The raters were experienced SLPs working separately. The procedure used consisted of two steps. In the first step, the raters decided whether the resonance was within normal limits or not. If it was not within normal limits, a rating of the degree of hypernasality was made on a three-point scale (mild, moderate or severe degree of hypernasality). The speech material on which the ratings were based had been coded and randomised so that the raters would be as blind as possible with respect to whether a child was IA or not. The score for each child was calculated as the median score of the ratings by the three raters. This score was then combined with scores for the number of active non-oral errors and symptoms of VP incompetence in the phonetic transcriptions to form the composite VPC-Sum score (Table 6).

In Study III, VP competence was analysed using VPC-Rate based on perceptual ratings (Lohmander et al., 2017c). For T1, samples of spontaneous speech were used and combined into a .wav file. For T2 and T3, recordings of the SVANTE sentences were used. For each child, there was one .wav file for each time point intended for perceptual analysis of VPC. All files were coded and randomised in order to make the judges blind as to timepoints as well. Ratings of overall perceived VP competence were performed by three experienced SLPs separately, on a three-point ordinal scale: competent (0), marginally competent (1) and incompetent (2) VP function. Marginally incompetent VP function was defined as existing when there was evidence of minor problems suggesting borderline VP closure not requiring surgery. Incompetent VP function was defined as existing when there was clear evidence of problems with VP closure that would require surgical intervention. The definitions were taken from Lohmander et al. (2017c). Finally, the rating chosen by the majority of the three judges for a child was reported as the VPC-Rate score for that child. For all analyses, 30% of the .wav files were doubled for the purpose of assessing inter and intra-rater agreement. Those files were also coded and presented in randomised order together with the other .wav files.

VP competence was not investigated in Study IV.

Table 6. Description and interpretation of the velopharyngeal-competence composite score (VPC-Sum)

Variables	*Definition*	*Composite score*		
		Score = 0	*Score = 1*	*Score = 2*
Sum of active non-oral errors from phonetic transcriptions	- glottal/pharyngeal plosives/fricatives - active nasal fricatives	0–2 errors	3–5 errors	>6 errors
Sum of the number of symptoms of VP incompetence from phonetic transcriptions	- audible nasal air leakage (nasal emission and nasal turbulence) - weak pressure consonants - nasal realisations of oral voiced consonants	0–2 symptoms	3–5 symptoms	>6 symptoms
Rating of hypernasality	Rated in a two-step procedure using a four-point scale (0–3)	Rated as 0	Rated as 1	Rated as 2–3
Sum of composite scores	The three variable scores are added up to calculate a VPC-Sum score with a possible range of 0–6			

	Interpretation of VPC-Sum		
	0–1	*2–3*	*4–6*
Definition	Competent VP function	Marginally incompetent VP function Evidence of minor problems suggesting borderline VP closure. Surgery is not required.	Incompetent VP function Evidence of significant problems with VP closure. Surgery is required.

Note: The numbers for the top two variables relate to the number of instances (not types) of active non-oral errors and the number of instances of manifestation (not types) of symptoms of VP incompetence, respectively. Abbreviation: VP = velopharyngeal.

Language material and analysis

Previous studies in cleft palate research have focused mainly on associations between speech production and expressive language. In this thesis, by contrast, it was decided to study ‘overall’ language ability, but to separate expressive language from receptive language ability in the tests and analyses – although the author acknowledges that the two are closely related and difficult to tease apart. Hence the assessment of receptive language was also considered important in the design of the project. In Study III, only expressive language ability was investigated; in that study, a specially designed questionnaire was also used to capture pre-adoption speech and language status as rated by the parents. In Study IV, both expressive and receptive language ability was investigated using standardised assessments as well as by means of a validated parental questionnaire for rating speech, language and communicative aspects.

Expressive language

In Studies III and IV, expressive language ability was assessed using:

Recalling Sentences, a subtest from the Clinical Evaluation of Language Fundamentals, fourth edition (CELF-4) (Semel et al., 2013). This subtest involves a repetition task where the child is instructed to repeat sentences with increasing length and complexity as correctly as possible, not changing the syntax or morphology of the sentence. The speech material produced by each child was transcribed orthographically and then scored by the assessor (the author). The results yield raw scores that are converted into scaled scores based on normative values (mean = 10, SD = 3). The Swedish version of the CELF-4 test, including the normative values, is based on 600 Scandinavian children aged 5:0 years to 12:11 years. Cronbach's alpha for this subtest is reported to be .89.

The *Bus Story Test* (BST) (Renfrew, 2002). In the BST, the test leader first reads a short story about a naughty bus to the child while showing a series of related pictures. The child is then asked to retell the story to the test leader using the pictures as support. The children's stories from the BST assessment were orthographically transcribed and then analysed by two final-year SLP students who worked independently and each transcribed approximately 50% of the audio recordings. The assessments relating to the three measures of Sentence Length, Information Score and Subordinate Clauses were also performed by the SLP students and in accordance with the manual. Sentence Length was calculated as the average length of the five longest sentences in each text. Information Score refers to the amount of information or story content that the child is able to retell, but it also reflects the child's ability to use his or her knowledge of syntax and morphology. In addition, Information Score also reflects how well the child is able to relate the main concepts of the story, whether he or she can present them in correct order and whether he or she can use the correct referent. Subordinate Clauses was calculated as the number of subordinate clauses. Each measure yielded a raw score which was compared with Swedish reference values.

All assessments were audio-recorded and scored in accordance with the respective test manual.

Receptive language

In Study IV, receptive language was assessed using:

The *Test for Reception of Grammar, version 2* (TROG-2; Bishop, 2009). The TROG-2 is a test of receptive grammar at the sentence level and is often used for clinical and research purposes. It is a picture-based test in which the test leader gives the child verbal instructions with increasing syntactic complexity to assess comprehension of grammatical structures. The child's task is to identify the picture, among four pictures shown, that matches the verbal instructions. The test includes 20 sets of different morphological and syntactic structures/constructions. Each set contains four tasks with verbal instructions pertaining to the specific structure/construction. Scoring was performed in accordance with the test manual. The test results are presented as raw scores, which can be converted to standard scores using normative values (mean = 100, SD = 15). The Swedish version includes normative values based on 650 children aged 4:0 years to 12:11 years and Cronbach's alpha is reported to be .89.

Parent-rated speech and language outcomes

Two parental questionnaires were used, one in Study III and another in Study IV. Both questionnaires were answered by one or two parents who accompanied the child at the time of speech and language assessment.

In Study III, a parental questionnaire designed to capture pre-adoption speech and language status was used. This questionnaire was produced specially for the present project, and it was also used to collect certain background variables described in Studies III and IV, such as the highest level of education in the family and the child's hearing status.

In Study IV, speech, language and communication were rated by parents using the Children's Communication Checklist – Second Edition (CCC-2; Bishop, 2012). The CCC-2 consists of 70 statements and the parent(s) rate how often each statement is true in the child's everyday life. The Swedish version of the CCC-2 has normative values for Scandinavian children aged 4:0 years to 16:11 years. There are ten different scales with seven statements in each. The scales are Speech, Syntax, Semantics, Coherence, Initiation, Stereotyped language, Use of context, Non-verbal communicative ability, Social relations and Interests. The last two scales, Social relations and Interests, were not included in the analyses in the present thesis. The results from the rated scales can be converted into scale scores for each scale, based on normative values (mean = 10, SD = 3). Cronbach's alpha (calculated on norm data for ages 4:9–12:11 years) for all scales was .78 (range: .69–.88).

Reliability/agreement

Prior to all assessments, transcriptions and ratings, training was carried out on material not included in the thesis for agreement purposes.

Speech production

In all studies, inter- and intra-transcriber agreement for the phonetic transcriptions was calculated as the percentage of agreement, point by point, and 30% of the speech material for which agreement was to be measured was duplicated and randomly presented alongside 'primary' speech material to measure intra-transcriber agreement. The author was the master transcriber of the phonetic transcriptions in Studies I–III and transcribed all speech material. Another SLP transcribed the duplicated 30% to enable measurement of inter-transcriber agreement. In Study IV, the phonetic transcriptions of all children plus transcriptions from the duplicated files were used to enable measurements of agreement.

Agreement for ratings of VP competence and hypernasality was calculated using different methods. In Study I, a binary scale rating of overall perceived VP competence was performed and then the percentage of agreement, point by point, was calculated on the basis of those ratings (see Table 7). Median percentages of inter- and intra-rater agreement were reported as results. In Study II, agreement was also calculated as the percentage of agreement, point by point, in the three raters' ratings of hypernasality. For intra-rater agreement, the percentage of agreement was reported. For inter-rater agreement, results were assessed on the basis of the frequency of (i) agreement between all three raters, (ii) agreement between two raters and (iii) no agreement at all. In Study III, where VPC-Rate was used for the perceptual assessment of overall VP competence, agreement was calculated in the same way as for the ratings of hypernasality in Study II.

Results from the agreement measurements relating to phonetic transcriptions and ratings are shown in Table 7.

Language ability

Agreement was measured for the assessments of expressive language based on the BST transcriptions in Study III. Two final-year SLP students both performed all assessments of all transcriptions of the children's retelling of the story separately. Inter-rater agreement for the three BST measures was calculated using Pearson's correlation coefficient. The values found for Sentence Length, Information Score and Subordinate Clauses were $r = .796$, $r = .905$ and $r = .630$, respectively. Two weeks after the first assessment, 30% of the transcribed material was reassessed to enable measurement of intra-rater agreement, also using Pearson's correlation coefficient. The values found for Sentence Length, Information Score and Subordinate Clauses were $r = .970$, $r = .979$, $r = .994$, respectively.

Statistical analysis

Non-parametrical statistical methods were used for statistical analysis in all studies. This choice was made not only because the samples were small but also because the data were skewed and not normally distributed. Group comparisons on independent samples were performed using the Mann–Whitney U test across all studies. Fisher's exact test was used on categorical data. In Study II, the VPC-Sum outcome was divided into three categories and group comparisons were then analysed using a χ^2 test.

For comparisons between dependent variables (Study III), the Wilcoxon signed-rank test was used to calculate differences in speech production between different timepoints. This choice was made because the samples differed in the number of children included at the different timepoints owing to missing data at T1. Hence comparisons were made between T1 and T2 and between T2 and T3.

To test correlations between variables in Studies III and IV, the Spearman rank-correlation test was used. In Study IV, correlations were calculated for both groups (i.e. IA children with and without CLP) together, rather than separately for each group, because prior statistical analyses had found no significant differences between the groups for the language variables.

A difference was considered to be statistically significant at the level of $p < .05$ (two-tailed). Statistical calculations were performed using the IBM SPSS statistics software, versions 22 and 25.

Table 7. Inter- and intra-transcriber agreement for phonetic transcriptions and inter-and intra-rater agreement for ratings of velopharyngeal competence, presented as median percentages and ranges

Study	*Speech-production ability*	*Assessment method*		*Inter-judge agreement*	*Intra-judge agreement*
I	Consonant proficiency	Phonetic transcription of SVANTE words	Place of articulation	81 (58–91)	92.5 (71–100)
			Manner of articulation	91 (71–100)	97 (71–100)
			Symptoms of VP incompetence*	91 (72–100)	97.5 (72–100)
			Non-oral articulation	-	-
	VP competence	Scale rating (binary outcome) of spontaneous speech	Between all three pairs of raters	76–84	100, 100, 100
II	Consonant proficiency	Phonetic transcription of SVANTE words	Place of articulation	81.5 (59–100)	93 (81–100)
			Manner of articulation	95.5 (66–100)	98 (81–100)
			Non-oral articulation	89 (75–100)	98 (86–100)
	VP competence	Phonetic transcription of SVANTE words	Symptoms of VP incompetence	77 (45–100)	87 (77–100)
		Scale rating of hypernasality (0–3) of the nine first SVANTE words	Agreement between all three raters (%)	31	Rater 1 53.8
			Agreement between two raters but not all three (%)	53	Rater 2 69.2
			All three disagree (%)	16	Rater 3 92.3
III	Consonant proficiency	Phonetic transcription of SVANTE words	Agreement on place and manner of articulation T1	81–91	92–97
			T2	81–96	93–98
			T3	85–97	91–98
	VP competence	Scale rating VPC Rate (0–2)	Agreement between all three raters (%)	64.3	Rater 1 64.3
			Agreement between two raters but not all three (%)	35.7	Rater 2 92.9
			All three disagree (%)	0	Rater 3 100
IV	Consonant proficiency	Phonetic transcription of SVANTE words	Agreement on voicing, place and manner of articulation	92	Rater 1 97
					Rater 2 97

Note: * Only audible nasal air leakage was included as a symptom of VP incompetence in Study I.
Abbreviations: SVANTE = Swedish Articulation and Nasality Test, VP = velopharyngeal.

Results

Speech production

Consonant proficiency

In all studies and at all ages, the IA children scored low on consonant proficiency and PCC (Figure 3). They differed significantly from the comparison groups across all studies. Although their PCC scores improved by age, the gap between their scores and normative data was large on a group level. In Study I, the IA children had a median PCC (and median PCC-A) score of 27.5%, almost 4 SD below the normative value, while the NA children with UCLP had a median PCC-A of 55.5%. The difference between the IA and NA children was significant (z = -2.964, $p < .01$). Significant differences between the groups in Study I were also found for PCP (z = -2.584, $p < .01$) and PCM (z = -3.585, $p < .001$).

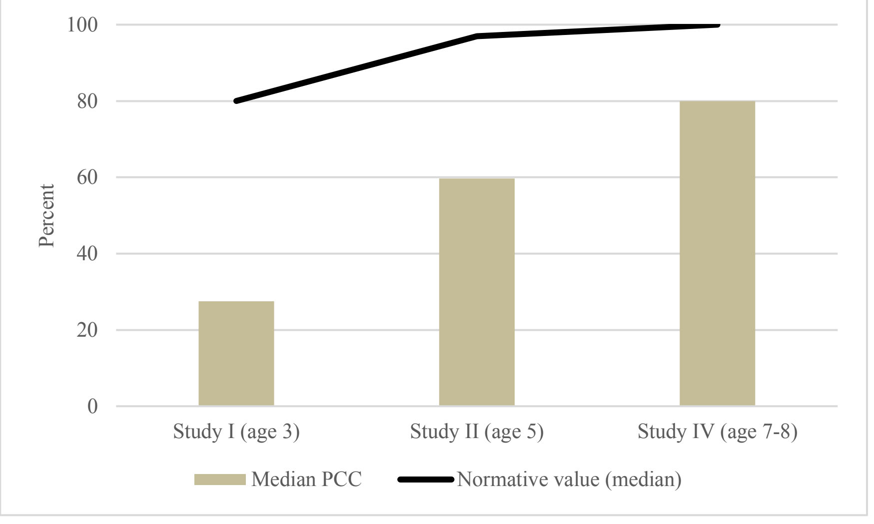

Figure 3. Overview of median PCC values for the internationally adopted children with unilateral cleft lip and palate in comparison with normative values (Lohmander et al., 2017).

At the age of 5 years (Study II), the IA children's median PCC score (59.7%) was 7.8 SD below the normative value. The IA children differed significantly on all PCC values from their NA peers with the same cleft type ($p < .05$). In addition, there was a large proportion of IA children with a PCC score of less than 50% (40% vs. 10% of the NA children; $p < .05$).

When the IA children with UCLP were 7 or 8 years old (Study IV) their median PCC (79.7%) was 6.1 SD below the normative value. While it should be noted that their PCC scores manifest a great deal of variability across all ages, it is worth pointing out that even at the age of 7 or 8 years, more than 80% of them scored 2 SD or more below the normative PCC value. This implies that many of the IA children with UCLP had substantial speech difficulties at school age.

In Studies I and II (at the ages of 3 and 5 years, respectively), the IA children with UCLP also had significantly lower scores than their NA peers with the same cleft type for other measures of articulation proficiency, i.e. PCP and PCM. Full results for the IA children with UCLP from each study with respect to consonant proficiency and consonant inventory are shown in Table 8.

In Study III, no significant correlations were found, at any timepoint, between age at adoption and any of the speech-production variables.

Consonant inventory

The consonant inventories of the IA children also developed significantly from the age of 3 years to the age of 7–8 years (Study III), but even so their inventories at each age must be qualified as limited (Table 8). At the age of 3 years, the IA children had a mean inventory of 5.4 (SD 2.73). At the ages of 5 years and 7–8 years, mean inventories of 12.2 (SD 3.5) and 15.7 (SD 1.8), respectively, were found. This can be compared with normative data showing that 85% of 3- and 5-year olds are typically able to produce 13 and 16, respectively, of the 18 Swedish consonants. Compared with NA children with the same cleft type (Study II), the IA children had significantly smaller consonant inventories than their peers at the age of 5 years ($z = -3.3$, $p = .001$).

Table 8. Results of the internationally adopted children with unilateral cleft lip and palate from each study for consonant proficiency (percentages) and consonant inventory (number of consonants)

	Study I		*Study II*		*Study III*		*Study IV*	
	Median (min–max)	Mean (SD)	Median (min–max)	Mean (SD)	Median (min–max)	Mean (SD)	Median (min–max)	Mean (SD)
PCC	-	-	59.7 (12–97)	-	T1 15.8 T2 60 T3 79.7	-	-	78.7 (14)
PCC-A	30 (2–70)	-	67.2 (25–97)	-	-	-	-	-
PCP	27 (6–75)	-	70.7 (23–98)	-	-	-	-	-
PCM	45 (19–84)	-	70.7 (23–98)	-	-	-	-	-
Consonant inventory	-	-	12 (5–18)	12	-	T1 5.4 (2.7) T2 12.2 (3.5) T3 15.7 (1.8)	-	-

Abbreviations: PCC = percent consonants correct, PCC-A = percent consonants correct, adjusted for age, PCP = percent correct place of articulation, PCM = percent correct manner of articulation, T1 = timepoint 1, T2 = timepoint 2, T3 = timepoint 3.

Consonant errors

Consonant errors were analysed in Studies I–III. The number of consistent consonant errors was established and the proportion of children with a consistent consonant error was reported.

At the age of 3 years (Study I), the IA children had a significantly higher frequency of glottal stops and glottal fricatives than their NA peers with the same cleft type ($p < .05$). Among the IA children, 78.6% used glottal stops and fricatives, as opposed to 27.8% in the comparison group. No other significant difference was found between the groups when it came to errors related to place of articulation. However, a high percentage of both IA (57%) and NA (83%) children with UCLP had oral retracted articulation as well. Further, 50% of the IA children as opposed to 22% of their NA peers had a consonant error defined as changing a plosive consonant into a nasal, although this was not a significant difference ($p > .20$). Deletion of target consonants was also very frequent in the IA children (78.6%), and this represents a significant difference ($p < .05$) relative to the comparison group (33.3%).

At the age of 5 years (Study II), cleft-speech characteristics (CSCs) occurred more frequently in the IA children (84%) than in the NA children (50%) ($p < .05$). Non-oral articulation, as an overarching category, was also more common in the IA children (44%) than in the NA children (25%), although this difference was not significant (p = .22). However, separate analysis of one type of non-oral consonant error, namely glottal articulation, showed that this was more frequent among the IA children (IA: 40%, NA: 5%; p = .012). The CSC of oral retracted articulation was a common error type in both groups (IA: 64%, NA: 35%) and there was no significant difference between the groups on that point.

Further, still at the age of 5 (Study II), developmental speech characteristics (DSCs) were common in both IA and NA children with UCLP. At least one type of DSC was found in the speech of 92% of the IA children and 65% of the NA children (p = .057). The DSCs of velar fronting (IA: 36%, NA: 5%; p < .05) and stopping of fricatives (IA: 28%, NA: 5%; p = .059) were both more frequent in the IA children than in the NA children. Difficulties with voicing represented a common error type in both groups (IA: 76%, NA: 55%; p = .21). Further, 40% of the IA children but only 10% of the NA children deleted target consonants; this was a significant difference (p < .05).

The results from Study III showed that the frequency of consonant errors decreased substantially with age (Table 9), although only some of the differences seen between timepoints were significant.

Table 9. Frequency of consonant errors at different timepoints for the internationally adopted children with unilateral cleft lip and palate in Study III

	T1		*T2*		*T3*		*Significance*	
	Age 3 years (n=13)		*Age 5 years (n=17)*		*Age 7–8 years (n=17)*			
	Median (min–max)		*Median (min–max)*		*Median (min–max)*			
Non-oral articulation	22.2	(0–52.9)	1.7	(0–21.1)	0	(0–18.3)	T1–T2 T2–T3	z = -2.9, p < .01 z = -2.1, p < .05
Oral retracted articulation	8.3	(0–70.4)	6.8	(0–35.6)	11.9	(0–28.8)	T1–T2 T2–T3	z = -1.7, p = .10 z = -0.4, p = .70
Deletion of target consonants	15.8	(0–56.1)	8.8	(0–53.6)	0	(0–10.3)	T1–T2 T2–T3	z = -2.9, p < .01 z = -2.3, p < .05
Developmental speech characteristics	17.5	(9.3–40.7)	12.3	(3.4–43.1)	3.5	(0–15.3)	T1–T2 T2–T3	z = -1.4, p = .20 z = -3.6, p < .01

Abbreviations: T1 = timepoint 1, T2 = timepoint 2, T3 = timepoint 3.

Velopharyngeal competence

Velopharyngeal (VP) competence was assessed in different ways across Studies I–III, but all assessments included perceptual ratings made by experienced SLPs.

Perceived VP increased by age (Figure 4). At the age of 3 years (Study I), 91% of the IA children with UCLP were perceived to have an incompetent VP function while this was the case for 35% of the children in the comparison group (p <.01). At the age of 5 years (Study II), many of the IA children (52%) were still perceived as having an incompetent VP function (defined as a VPC-Sum score of 4–6), but at that age they did not differ significantly (p = .17) from their NA peers (for whom the corresponding proportion was 25%). It should be noted that at the age of 5, four of the IA children (and two of the NA children) had undergone secondary VP surgery prior to the speech assessment. All of those four IA children were deemed to have an incompetent VP function even after their secondary surgery.

In Study III, although improvement in VP competence was seen between the age of 3 years and the age of 7–8 years, 18% (n=3) of the IA children with UCLP were still rated as having an incompetent VP function at the age of 7–8. Moreover, 41% of them (n=7) had a marginally incompetent VP function at that age while only 41% (n=7) had a competent VP function. Six IA children (35%) had received secondary VP surgery prior to the speech assessment performed at the age of 7–8 years, but only one of those six was deemed to have a competent VP function at the time of that assessment while the other five were rated as having a marginally incompetent (n=3) or incompetent (n=2) VP function.

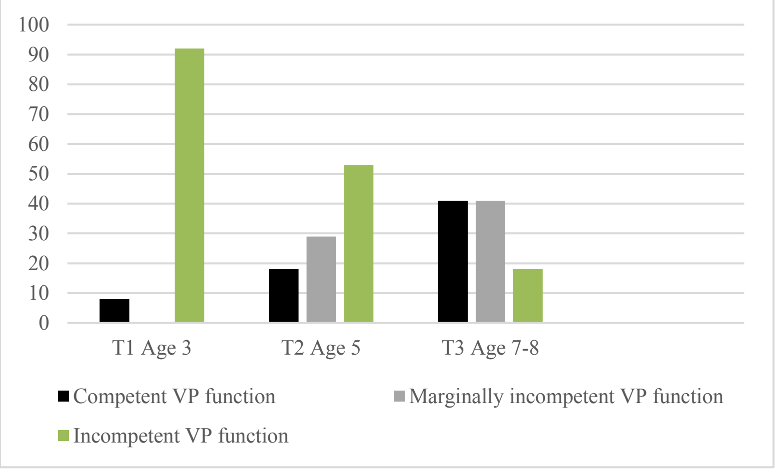

Figure 4. Results based on VPC-Rate at different timepoints and ages, from Study III.

Language ability

Expressive language ability

The scores for expressive language ability at the age of 7–8 years (Studies III and IV) were in the lower average range at group level.

On the Recalling Sentences task in Study III, the IA children with UCLP obtained a mean scaled score of 7.4 (SD 3.3). A total of 41% (n=7) children scored more than 1.25 SD below the age-specific norm. Three children (18% of the entire group) scored more than 2 SD below that norm. On the Bus Story Test, in Study III, the group obtained a mean SL raw score of 8.8 words per sentence (SD 2.1). This value corresponds to the norm for children aged 6:1 to 6:6 years as indicated in the Swedish BST reference values. At the same age as the children in Study III (7–8 years), Swedish children typically have a mean Sentence Length score of 11. The mean Information Score of the children in the present study was 22 (SD 9.4), which corresponds to the norm for the 4:0-4:11 age range. The typical score for children at their age is 33 (SD 6.87). Finally, the mean Subordinate Clauses score for the IA children with UCLP in Study III was 2.5 (SD 1.7), which corresponds to the norm for the 4:4-5:0 age range. The typical mean Subordinate Clauses score for children at the age of 7–8 years is 4.0.

In Study IV, no difference was found between the IA children with UCLP (7.6 SD 3.0) and those without UCLP (6.9, SD 2.3) (z = -.88, p = .38) on the Recalling Sentences task, indicating that the presence of CLP did not have an impact on expressive language development. However, if the IA children's scores are compared with test norms and reference values from Swedish children, a large proportion of them scored poorly on all measures of expressive language. In Study IV, almost 15% (n=4) of the IA children with UCLP and 14% (n=4) of those without UCLP scored at least 2 SD below the normative mean on the CELF-4 subtest of Recalling Sentences. When it comes to the expressive language measures based on the BST, both IA groups scored at a level typical of NA children approximately two years their juniors, and the groups did not differ significantly from each other on this point either. For Information Score, both groups had particularly weak results, obtaining values typical of NA children aged 2.5 years less. Table 10 shows the scores on each measure of expressive language for the two groups in Study IV.

Table 10. Scores on measures of expressive language for the two groups in Study IV

			IA +UCLP n = 27	IA Non-UCLP n = 29	Significance z-value	p-value
Expressive language	CELF-4 (sub-test)	Recalling Sentences, mean scale score (SD)	7.6 (3.0)	6.9 (2.3)	-0.88	$p = 0.38$
	BST	Sentence Length, mean raw score (SD)	9.3 (2.4)	9.4 (2.1)	-0.30	$p = 0.76$
		Information, mean raw score (SD)	24.2 (9.5)	23.0 (7.8)	-0.51	$p = 0.61$
		Subordinate Clauses, mean raw score (SD)	2.9 (1.8)	2.7 (1.7)	-0.27	$p = 0.79$

Notes: The Mann–Whitney U-test was used to analyse differences between groups. Abbreviations: IA = internationally adopted, UCLP = unilateral cleft lip and palate, CELF-4 = Clinical Evaluation of Language Fundamentals – version 4, BST = Bus Story Test

In Study III, there were no significant correlations between age at adoption and any of the expressive- language variables. In Study IV, correlations between age at adoption, time of exposure to Swedish and expressive language ability were calculated. The BST measures of Sentence Length and Subordinate Clauses correlated weakly, though significantly, with age at adoption (Sentence Length: $r_s = -.35$, $p < .01$; Subordinate Clauses: $r_s = - .27$, $p < .05$) and with exposure time (Sentence Length: $r_s = .36$, $p < .01$; Subordinate Clauses: $r_s = .32$, $p < .05$).

Receptive language ability

The results for receptive language in Study IV did not differ significantly between IA children with UCLP (mean standard score 90.5, SD 14) and IA children without UCLP (mean standard score 94, SD 13.4) ($z =-.93$, $p = .35$); at group level, both groups scored within the average range. When it comes to the prevalence of impairment, the result depends on which cut-off is used (-2SD or -1.25 SD). A score of at least 1.25 SD below the normative mean was found for nine (33%) of the IA children with UCLP but for five (17%) of the IA children without UCLP. By contrast, very low scores for receptive language of 2 SD or more below the norm were found for two (7%) of the IA children with UCLP but for no child in the comparison group.

However, no significant differences were found between the two groups in terms of the prevalence of impairment regardless of the cut-off used.

Weak correlations were found between receptive-language scores and age at adoption (r_s = -.29, $p < .05$) and exposure time (r_s = .33, $p < .05$), respectively.

Parental perspectives on speech, language and communication

In Study III, parents were asked to report on their children's pre-adoption speech and language status using a simple questionnaire, and an attempt was then made to explore and relate the children's speech and language development at the ages of 3, 5 and 7–8 years to their pre-adoption status.

Most children (47%) in Study III were reported to have used babbling before the adoption. Further, 12% were reported not to have had any speech at all, while 12% were said to have used words and 6% (one child) sentences in their native language (Table 11).

The description in Study III of the relationships between parent-reported pre-adoption speech and language status and expressive language ability at the age of 7–8 years was based on a small overall sample, meaning that the subgroups identified were even smaller. For this reason, no statistical calculations were performed on the potential relationships. However, interestingly, the children who had used words and sentences at the time of adoption did obtain higher scores at the age of 7–8 years than those who had been silent or just used babbling at the time of their adoption (Table 11).

In Study IV, parental ratings using the CCC-2 questionnaire showed that the only aspect that differed between the groups of IA children with and without UCLP was speech ability, for which the parents of IA children with UCLP scored their children's speech significantly lower (z = -2.2, $p < .05$). Around 20–30% of the IA children with and without UCLP scored 1.25 SD or more below normative mean scaled scores for several subscales, implying that many children were at risk of having a language disorder (Table 12).

Table 11. Pre-adoption speech and language status as reported by parents in relation to speech-production scores at different ages and expressive language scores at the age of 7–8 years in 17 children (Study III)

Pre-adoption speech and language status	*Number of children (%)*	*PCC T1*	*PCC T2*	*PCC T3*	*VPC-Rate T1*	*VPC-Rate T2*	*VPC-Rate T3*	*Recalling Sentences (scale scores) T3*	*BST: SL/Inf/Sub clauses (raw scores) T3*		
No babbling or speech at all	2 (12)	3.7	45.1	80.6	2	1	0	7	8.8	22.5	2.0
Babbling	8 (47)	14.8	63.6	82.3	2	2	1	7	8.7	23.8	2.5
Use of words in native language	2 (12)	15.8	62.8	76.1	2	1.5	1	9	9.2	25.5	2.5
Use of sentences in native language	1 (6)	56.9	72.4	86.4	0	0	0	11	10.8	29.0	3.0
Do not know	4 (23)	22.9	49.5	71.6	2	1.5	1	7	8.2	14.8	2.8

Note. PCC and VPC-Rate are reported as median values. Recalling Sentences and BST scores are reported as mean values. Abbreviations: PCC = percent consonants correct, T1 = timepoint 1, T2 = timepoint 2, T3 = timepoint 3, VPC-Rate = velopharyngeal-competence rate, BST = Bus Story Test, SL = Sentence Length, Inf = Information Score, Subclauses = Subordinate Clauses.

Table 12. Number (%) of children in each group obtaining scaled scores at least 1.25 SD and 2 SD, respectively, below the normative mean on the CCC-2 subscales

	IA +UCLP n = 27 (%)		*IA Non-UCLP n = 29 (%)*		*Significance*	
	-1.25 SD	-2 SD	-1.25 SD	-2 SD	-1.25 SD	-2 SD
Speech	17 (63)	11 (40.7)	8 (27.6)	6 (20.7)	$p < .05$	$p = .15$
Syntax	9 (33.3)	2 (7.4)	8 (27.6)	3 (10.3)	$p = .77$	$p = 1.0$
Semantics	5 (18.5)	1 (3.7)	3 (10.3)	1 (3.4)	$p = .46$	$p = 1.0$
Coherence	9 (33.3)	2 (7.4)	4 (13.8)	3 (10.3)	$p = .12$	$p = 1.0$
Initiation	7 (25.9)	2 (7.4)	6 (20.7)	1 (3.4)	$p = .76$	$p = .61$
Stereotyped language	6 (22.2)	0 (0.0)	4 (13.8)	1 (3.4)	$p = .50$	$p = 1.0$
Use of context	6 (22.2)	1 (3.7)	9 (31)	1 (3.4)	$p = .56$	$p = 1.0$
Non-verbal communicative ability	1 (3.7)	0 (0.0)	6 (20.7)	2 (6.9)	$p = .10$	$p = .49$

Abbreviations: IA = internationally adopted, UCLP = unilateral cleft lip and palate, CCC-2 = Children's Communication Checklist – second version.

In summary, the results for speech and language ability at the age of 7–8 years, as measured using scores on standardised speech and language tests and parental ratings, showed that the IA children with UCLP obtained scores comparable to those of the IA children without UCLP. The only variable that differed significantly between the groups concerned speech ability – both according to the speech measure (PCC) and according to the parental rating. However, large proportions of the children in both groups obtained low scores for the various measures of expressive language ability. Only weak correlations were found between the measures of receptive and expressive language, on the one hand, and age at adoption and time of exposure to Swedish, respectively, on the other hand.

Discussion

The overall aim of the present thesis was to investigate speech-production development from a longitudinal perspective in internationally adopted (IA) children with unilateral cleft lip and palate (UCLP) originating from China. A further aim was to investigate those children's language ability at the age of 7–8 years. This was done by assessing the IA children's speech production in detail at different ages, by comparing their proficiency with that of different groups of same-age peers and by exploring and describing potential associations between pre-adoption speech and language status, age at adoption and time of exposure to Swedish. Parental ratings of speech, language and communication aspects were also included.

In summary, the results showed that although the IA children had significantly lower articulation proficiency at the ages of 3 and 5 years than NA children with the same cleft type, the IA children made substantial progress in terms of articulation and perceived velopharyngeal competence up to the age of 7–8 years. However, it was also found that a majority of the IA children with UCLP still had persistent difficulties with speech production even at the age of 7–8 years and few had age-appropriate speech. Interestingly, it was found that a high proportion of IA children with *and* without UCLP had expressive language difficulties at the age of 7–8 years, which suggests that being internationally adopted may in and of itself entail a risk of delayed language development for many years after the adoption.

Internationally adopted children with UCLP have been treated as a single group in the present thesis, but it is important to acknowledge the heterogeneity found within this group. Assessing speech and language development in *any* child is a complex task, especially in young pre-school children, whose ability to participate varies widely. Investigating speech and language development in IA children is an even greater challenge, since there are numerous variables which are impossible to control for. For example, in the case of most IA children, there are several medical and psychological pre-adoption factors and circumstances of which little or nothing is known and which may or may not influence later development. Despite this, many researchers before us have performed investigations on IA children and their speech and language development, and we have continued to do so.

In an attempt to make a heterogeneous group somewhat less heterogeneous, we chose to include only children originating from China and having a specific cleft type who were adopted during the 'peak' of international adoptions of children with cleft lip and palate. Additionally, in two of the studies, only children treated by a specific CLP team during the same period of time were included as participants, both in the study group and in the comparison group. A second comparison group consisting of IA children without cleft lip and palate was also used. While it is true that those children had been adopted from different countries, at least they were adopted during the same time period and into families with similar educational levels. In fact, at the outset of the present PhD project, the intention was to include only children born in China in the comparison group without cleft lip and palate. Although many languages and dialects are spoken in China, this could have increased the likelihood that all adopted children studied would have a more similar language background. Unfortunately, this proved impossible, because few of the children adopted from China at the time in question did not have a cleft lip and palate.

Study I suggested that at the age of 3 years, the IA children with UCLP had significantly lower consonant proficiency and velopharyngeal competence than NA children with the same cleft type. Although that study was one of the very first to perform a detailed investigation of speech production in IA children with CLP, this was an expected finding. At that time, the IA children had been adopted quite recently, they had in most cases recently undergone surgery for palatal closure, and some of them still had an open hard palate or large palatal fistulae. It was decided to include only those children who had produced sufficient speech material in order to enhance the likelihood of a fair assessment and analysis. Hence a cut-off of ≥50% target consonants produced was used as an inclusion criterion, meaning that, to be included in the analysis, a child had to have named at least 50% of the target words in the SVANTE test during her or his speech assessment. Application of this criterion entailed the exclusion of eight IA children and three NA children. The fact that so many IA children had to be excluded for this reason was interpreted as a symptom of the prevalence of more severe speech and language difficulties among the excluded children, but in fact this could also have reflected the postponement of those children's language-acquisition process, which may have caused them not to have acquired enough Swedish at the time of assessment. In addition, other factors may also have had an impact on outcomes in terms of speech production and on the differences found between the groups. For example, there was a great difference in the timing of palatal repair. While the IA children underwent soft-palate repair at a mean age of 24 months, the NA children did so at a mean age of 6 months. Previous retrospective studies of IA children with CLP (e.g., Sullivan et al., 2014) have also described a higher prevalence of VP incompetence compared with groups of NA children with CLP, suggesting that the timing of palatal repair and the period spent with adequate VP closure (Morgan et al., 2018) may be linked to speech proficiency. Although the present thesis included a detailed analysis of speech production, its results need to be interpreted with caution since the sample studied was small and since there was a great deal of variety within the group of IA children. A second factor that may have influenced speech outcome is differences in hearing thresholds and/or ear status between the IA children and the NA children. The NA children in the present thesis had had their ear and hearing status assessed regularly since birth and most likely also had had pressure-equalising tubes inserted or received other early hearing interventions as needed, which it is unlikely that the IA children received at such an early age. However, the children in Study I did not differ in terms of pure-tone averages (PTAs) at the time of speech assessment. In fact, more children in the NA group than in the IA group were considered to have a hearing loss (defined as PTA > 25dB) on one or both ears. Other researchers have found similar results, i.e. no difference between IA children with CLP and NA children with CLP in terms of the total number of episodes of otitis media with effusion during childhood, in terms of hearing thresholds (PTAs) at the age of 2–6 years (Werker et al., 2018) or in terms of the prevalence of hearing loss at the age of 5 years (Sahlsten Schölin et al., 2020).

At the age of 5 years (Study II), the IA children continued to manifest lower scores on consonant proficiency for all variables compared with same-age NA peers with same cleft type. Like at the age of 3 years, the results varied greatly; it is obvious that there are large differences within the group of IA children. As 5-year-olds, a few IA children scored in line with normative values, but most of them did not. Of the 25 IA children, 10 had a PCC score of less than 50%, which is far behind NA children with UCLP and NA children without cleft lip and palate. In fact, a PCC score of less than 50% is even below the normative value for Swedish 3-year-olds without cleft lip and palate (Lohmander et al., 2017a). One interesting finding when it comes to developments between the ages of 3 and 5 years is that the IA children made great advances in terms of PCM (percent correct manner of articulation). At the age of 3 years (Study I), their average PCM was 45% (min–max 19–84), but at the age of 5 years (Study II), this value had increased to 92.7% (min–max 28–100). This could be due to progress in language development, but it could also reflect the fact that more children had complete closed palates and adequate VP competence. At the age of 3 years, five IA children still had an open hard palate owing to residual clefts or fistulae. At the age of 5 years, four of them had undergone secondary VP surgery. Although many (about half) of the IA children were still rated as having incompetent VP function, this – including the fact that almost all of the children had spent longer, and relatively speaking much longer, with a functional palate – could have affected the outcome and improvement seen in terms of PCM.

Further, PCP (percent correct place of articulation) had also improved a great deal at the age of 5 years. Even so, average PCP scores were lowered by the fact that many children (IA: 64%, NA: 35%) made a great many errors related to place of articulation (oral retracted articulation) at the age of 5 years. Here it should be noted that although some children had undergone surgical procedures relating to palatal fistulae at the age of 5 years, many of them still manifested a high frequency of oral retracted articulation. This type of articulation error is common in children with CLP (Willadsen et al., 2017) and seems to be particularly common in children who have had a residual cleft or fistula in the hard palate, i.e. even after this has been surgically corrected (Lohmander et al., 2006).

Despite the low overall level of PCC scores, however, progress in consonant proficiency by age was significant (Study III). This positive development was probably affected by increases in the number of surgically closed palates, in age, in time after surgery and in the amount of secondary VP surgery. Several studies have described a higher frequency of secondary VP surgery in IA children than NA children with CLP (Morgan et al., 2018; Sahlsten Schölin et al., 2020) and it has also been suggested that the longer time a person spends with a sufficient VP function, the better her or his articulation skills will be (Morgan et al., 2018). Morgan et al. (2018) also reported weaker articulation skills in IA children with incompetent VP function. The close relationship between a competent VP function and proficient articulation is thus well known, and it was found in this thesis as well. However, more IA children than NA children had received secondary VP surgery and palatal-fistula repairs by the age of 5 years (Study II), but this did not seem to affect the differences in consonant proficiency.

Low PCC scores were also evident at the age of 7–8 years (Study IV), when the IA children's results were compared with normative data for the SVANTE and with the scores of the IA children without CLP. At the age of 7–8 years, most IA children with UCLP (80%) scored at least 2 SD below the normative mean. At that age, the IA children, on average, had had a complete palate for around 4 years. However, it is highly likely that most of them had not yet had their alveolar cleft closed, and this may have affected their consonant production. Some of the children in Study IV had had this surgical procedure performed prior to the speech assessment at the age of 7–8 years, but the majority of them had not.

In conclusion, our results for consonant proficiency suggest that there is a risk that IA children lag behind their NA peers without CLP and that there remains a wide gap between them at least until the age of 7–8 years. Many IA children present with low consonant proficiency at the age of 7–8 years, and in combination with VP incompetence (to different degrees), this places them at risk of reduced intelligibility, which may in turn affect their social communication.

Different types of consonant errors were found to occur frequently in both groups of children with UCLP, although some of them were more common among the IA children. This finding was in line with the study by Morgan et al. (2018). The studies in the present thesis found one characteristic type of non-oral articulation – glottal articulation – to be more common among the IA children than among their NA peers both at the age of 3 years (Study I) and at the age of 5 years (Study II). However, at the age of 7–8 years (Study III), the prevalence of glottal articulation had decreased significantly, although there was great variation. Glottal articulation is closely related to an incompetent VP function, which 18% of the children had at the age of 7–8 years (Study III). Another commonly occurring consonant error was oral retracted articulation, which was also more frequent in the IA children (64% vs. 35% in the NA children) at the age of 5 years (Study II), although the difference was not statistically significant. Interestingly, Study III showed an increase in the prevalence of oral retracted articulation in the IA children between the age of 5 years (6.8%) and the age of 7–8 years (11.9%), but that difference was not significant. Although oral retracted articulation is a common error in children who have undergone a two-stage palatal-repair procedure with delayed hard-palate repair (Lohmander et al., 2006), this finding was unexpected. The reason for this increase in the frequency of oral retracted articulation could be that the target consonants which were orally retracted at the age of 7–8 years had simply been missing at the age of 5 years. In fact, at the age of 5 years, most of those target consonants were either replaced by glottal stops and fricatives or entirely missing (i.e. deletions of target consonants).

The analysis of the consonant errors found at the age of 5 years (Study II) yielded a complex picture of the IA children's consonant production. The NA children's consonant-error patterns were more consistent while those of many IA children were more 'mixed up'. In other words, a mix of different error patterns appeared in many IA children's production in Study II and no such mix was evident for the NA children. Although the IA children had a high frequency of oral retracted articulation, many of them had established anterior consonants and also had a high frequency of the developmental speech characteristic of velar fronting. Morgan et al. (2018) found a high frequency of articulation impairment among their IA children aged 5–6 years as well. Additionally, at that age, they found a higher frequency among IA children than among NA children of cleft-related errors in combination with developmental errors, and this difference was even more evident at the age of 7–8 years. In Study II, a high frequency of developmental speech characteristics was found for both IA and NA children, but velar fronting and stopping stood out as more prevalent among the IA children. In Swedish children without CLP, these types of consonant errors, and also most other developmental speech characteristics, are rare at the age of 5 years. Hence it seems that certain types of developmental consonant errors are more frequent among IA children with CLP. It is of course only possible to speculate about the reason for this, but it might be due to the IA children's early babbling experiences, which were affected by their open palates as well as their pre-adoption language environment and the change of languages that they underwent at a time when language is developing a great deal. There seems to be a higher risk of inappropriate learning of consonants and a higher risk of phonological difficulties in IA children with CLP, and exploring the reasons for this clearly represents an important objective for future studies. A detailed investigation of phonological aspects, for example including tasks to assess phonological awareness and processing ability, would have yielded a great deal of important information that could have implications for whether IA children are successful in learning how to read as well as for what could be done to help them succeed at that task.

Perceived VP competence improved from the age of 3 years to the age of 7–8 years for the children with UCLP in this thesis. Previous studies of IA children with cleft lip and palate have all shown a high risk of incompetent VP function and a greater need for secondary VP surgery to improve speech (Follmar et al., 2015; Morgan et al., 2018; Sahlsten Schölin et al., 2020; Sullivan et al., 2014; Swanson et al., 2014). There are several factors that may explain the improvement of VP competence seen among the IA children in the present thesis. First, many children still had an open hard palate at the age of 3 years (n=5). By contrast, at the age of 5 years, most of them had better anatomical conditions, which probably resulted in better VP competence, although one IA child did have a large unoperated palatal fistula at the time of assessment. At the age of 7–8 years (Study III), approximately 40% of the IA children were rated as having a competent VP function. Among those children, one had received secondary VP surgery and palatal-fistula repair and another had undergone palatal-fistula repair prior to the age of 7–8 years. Further, three children were rated as having an incompetent VP function at the age of 7–8 years. Two of those had complicating factors such as repeated secondary VP surgery and palatal-fistula repair prior to the speech assessment. By contrast, the third child rated as having an incompetent VP function had not undergone any secondary VP surgery prior to the speech assessment at the age of 7–8 years.

In the present thesis, perceived VP function has been assessed using different perceptual assessments, in contrast to Morgan et al. (2018), who assessed VP function by measuring the VP gap using the PERCI speech aerodynamic assessment system. Further, the present thesis used different types of speech material: spontaneous speech (Study 1), repeated sentences (Study III) and single words (Study II). Continuous speech is thought to be more sensitive to VP incompetence, and also more similar to everyday communication. Hence assessment based only on single-word speech material risks underestimating a potential VP dysfunction. In Study I, VP competence was assessed on the basis of spontaneous speech because the children were so young that it would not have been possible to have them repeat sentences. Further, the available speech material for many children in Study I was scant, in all likelihood because many of them had not been exposed to Swedish for a very long time. However, the perceptual assessment made in Study I, using a rating scale with only two steps (competent vs. not competent) is rather broad and so risks yielding a fairly rough perceptual measure of VP competence. In Study II, at the age of 5 years, VPC-Sum, which is a composite score, was used to assess VP competence in greater detail. This score combined perceptual rating of hypernasality in a nine-word string with calculations of VP symptoms (nasal air flow and weak pressure consonants) and non-oral articulation errors from the phonetic transcriptions. VPC-Sum has been shown to be a valid and reliable method for this purpose and is recommended for use in research (Lohmander et al., 2017c). In Study III yet another method was used: VPC-Rate, where perceived VP competence is rated for continuous speech on an ordinal scale (0–2). A previous validation study showed significant positive correlations between VPC-Rate and – the more detailed and time-consuming – VPC-Sum (Lohmander, Hagberg, et al., 2017). Sahlsten Schölin et al. (2020) performed an overall perceptual rating using a three-point scale (the same VPC-Rate measure as used in Study III in the present study) and speech material consisting of sentences. That different methods for analysing perceived VP function were used in the present thesis could be seen as a limitation, and the results need to be interpreted with these differences in mind. However, all rating methods were chosen to suit the specific aim of each study, and they were all performed by three blinded, independent and trained SLPs.

The results for language ability suggest that there is an increased risk of delayed expressive language in the IA children who participated in the present thesis. While previous studies had raised concerns specific to IA children with cleft lip and palate (Morgan et al., 2017; Scherer et al., 2018), no previous study had compared IA children with and without cleft lip and palate. The present thesis actually did not identify any language abilities or difficulties that 'stood out' specifically for the children with cleft lip and palate. However, compared with test norms (and hence indirectly with NA children), both groups of IA children (with and without cleft lip and palate) in the present study manifested difficulties on all tasks of expressive language. Previous studies have found expressive language difficulties in school-aged IA children adopted from China (Delcenserie et al., 2013). The children in that study scored significantly lower than their NA peers (matched for age, gender and socioeconomic status) on measures of receptive grammar, expressive vocabulary, word definitions and sentence recall. The authors also found a significant correlation between all language scores (both receptive and expressive language) and the Recalling Sentences subtest of the CELF-4. Although our findings did not concur on measures of receptive language, the results of the present thesis (Study IV) in terms of poor performance on measures of expressive language are in line with Delcenserie et al. (2013). One important difference, though, is that the present thesis (Study IV) compared language outcome with test norms and unfortunately did not include comparison groups of non-adopted children with and without cleft lip and palate. The inclusion of such groups would have yielded more knowledge about language-development trajectories and enabled an even greater contribution to filling the knowledge gap when it comes to IA children with and without cleft lip and palate.

The task of repeating a sentence has been reported to be a measure of verbal memory (Alloway & Gathercole, 2005). However, previous research supports the idea that this task is in fact rather a measure of the child's overall language ability (Klem et al., 2015) and hence possible to use as a clinical marker in identifying language disorders (Vang Christensen, 2019). Although sentence repetition demands high attention skills and linguistic knowledge, it seems safe to say that it is also an appropriate method to assess expressive language ability. Morgan et al. (2017) performed a much broader test of language ability (with the Recalling Sentences subtest as one of several components). They found that 24% of the IA children with CLP (vs. 18% of the NA children with CLP) could be considered impaired in their expressive language, because they performed at or below a standard score of 80. In the present thesis (Study IV), the mean scaled score on Recalling Sentences was within the lower average range at group level, but 15% of the IA children with UCLP scored at least 2 SD below the norm on Recalling Sentences.

In Study IV, the results showed no significant difference between the two groups of IA children for the single measure of receptive language used. However, around 30% of the IA children with UCLP scored at least 1.25 SD below the normative mean, as did 17% of the IA children without UCLP. Further, two of the IA children with UCLP but no child in the comparison group scored very low (at least 2 SD below the norm). These differences between the groups were not significant. The high proportion of children scoring at least 1.25 SD below the norm was rather an unexpected finding, since no child had been reported to have a developmental language disorder and also given that, at this point, the mean time of exposure to Swedish in the group of IA children with UCLP was more than 5 years. In line with the results of the present thesis, Morgan et al. (2017) also found a high frequency of IA children with CLP (20%) deemed to have an impaired receptive language (at or below a standard score of 80).

Receptive language ability is made up of much more than just the grammar comprehension assessed with the single test that was used in Study IV. Receptive vocabulary and comprehension of narratives are also important functions of language ability. Unfortunately, those aspects were not included in the present thesis. It would be a good idea for future research to assess receptive language ability more broadly.

Further, the choice of language tests and measures can be questioned. It was decided to use tests with normed references for Swedish children wherever this was possible. In this context, it is relevant to ask what constitutes a valid and reliable language test beyond statistical aspects such as reasonable reliability in terms of, for example, internal consistency. There are several aspects to consider when assessing language ability in a child. For example, all tests of language ability used in the present thesis require the child to have a certain level of ability to focus and concentrate. Hence aspects such as attention, the ability to focus and concentrate on a task, verbal working memory, etc., need to be kept in mind. This is because difficulties in those areas may cause a child to score poorly on tests such as those used in the present thesis, meaning that the reason for the low scores will not necessarily be potential language difficulties. This was in fact taken into consideration in the design of the studies. The language tests used in the present thesis are tried and tested, because they are commonly used by Swedish SLPs when diagnosing language disorders in children from the age of 4 years. There is unfortunately a lack of valid and reliable test instruments that have Swedish normative data; the tests chosen for the present thesis are the ones with the most reliable comparison data. One potential problem to be kept in mind is that the Bus Story reference values are based on a fairly small number of Swedish children. Additionally, to ensure that the children participating in Study IV (where assessment took rather a long time) would have a fair chance of performing at their best on the language-assessment tasks, other considerations related to the test situation were also taken into account, for example to limit potential worries that the children might have or reduce any difficulties focusing on the task.

Study III explored pre-adoption speech and language status in relation to post-adoption speech-production and language-ability outcomes. A questionnaire was specially designed for this purpose. Hence the parents were asked to report on their child's pre-adoption status at a time when he or she was 7–8 years old. This can be questioned for many reasons. First, reporting information on an ability more than four years after the adoption can be difficult and there is a risk that parents may have over- or underestimated their child's past speech and language ability. Second, the parents were asked to report whether their child was using any babbling, words or sentences in his or her native language at the time of adoption. This is also a very difficult question to answer. Some parents do receive detailed medical records, including in some cases a speech and language assessment report from an SLP in the child's country of origin who saw the child before the adoption, and some parents had also received information from the carers at the child's orphanage. However, some parents had to estimate their child's pre-adoption speech and language status themselves, without any such support. In addition, most parents probably did not know their child's native language at all (making it difficult to distinguish babbling from speech). Further, even though almost all adopted children are accompanied by medical records at the time of adoption, those records differ immensely in many respects. Finally, many of the parents participating in the present thesis have pointed out that the information contained in those medical records seemed highly questionable given the findings made during post-adoption medical examinations in Sweden.

To take a broader perspective on speech and language ability, a validated questionnaire for parental rating – the CCC-2 – was used in Study IV. Parental perspectives on speech, language and communication are important to gain further insight into any potential difficulties that a child may have. Also, parental perspectives may help researchers and clinicians to gain a deeper knowledge of the potential impact that a speech and language impairment may exert on everyday life.

The CCC-2 questionnaire was originally designed as a tool to identify language disorders and specifically to identify impairments of pragmatic language in children manifesting disorders of communication. It was also designed to identify children with language disorders that may require further assessment for a potential autism-spectrum disorder. The CCC-2 is commonly used in Sweden and is one of few questionnaires of its kind with Swedish norms (from the age of 4 years to that of 16:11 years). The CCC-2 can be an effective way to measure aspects of speech and language that are not very easily captured at a clinic in formal testing or evident at the time of testing. It may also help the SLP obtain a more ecologically valid measure of speech and language ability in that the parent is asked to report strengths and difficulties that he or she has observed in daily life. Interestingly, there was only one aspect (out of eight) that differed significantly between the groups in Study IV, namely speech ability. However, there were individual children in both groups who were rated as having difficulties (at least 2 SD below the mean) within several aspects/subscales. One of the purposes of Study IV was to investigate whether IA children with and without CLP differed, at group level, in terms of parental ratings on speech, language and communication aspects. Overall, it turned out that they did not – but on closer examination of the results of the CCC-2, it is evident that there are children in both groups who are at a risk of having a language disorder.

Sample characteristics and sample size are common grounds invoked to question the findings and outcomes of research studies. In Studies I–III, all IA children with UCLP at a tertiary Swedish clinic were asked to participate, and a high proportion of families accepted the invitation to participate. Hence, although the samples were small, they represent a large proportion of the population of IA children with UCLP treated at that clinic. It was decided to include only children with UCLP, since this was the most common type of CLP at that clinic – and also the most common type in the earlier retrospective studies of IA children with CLP which were reviewed (Follmar et al., 2015; Goldstein et al., 2014; Hansson et al., 2012). The comparison group for Study IV was recruited through advertising. In a sense, this can be said to constitute a convenience sample, since all assessments were to be performed in the children's homes by the author. Advertising for participants can increase the risk of selection bias in that children with difficulties in language ability and children performing exceptionally well in terms of language ability may have parents who are more likely, for one reason or another, to sign up their child for participation. This was an apparent risk in the present thesis, and the results need to be interpreted with that in mind.

Studying relationships between different values can be difficult if the sample is small or if the participants included in the sample are highly similar or dissimilar. In such cases, any correlations calculated must be interpreted with caution. In Study III, no correlations at all were found between consonant-proficiency variables, age at adoption or expressive language. The only correlations found in that study were within each variable of consonant proficiency studied and between the different measures of expressive language. It had been expected that there would be a relationship between age at adoption and expressive language, since many prior studies had found such a link (Morgan et al., 2017; Scott et al., 2008). In addition, previous studies of NA children with cleft palate had also found a relationship between early speech production and later expressive language (Chapman et al., 2003; Scherer et al., 2008), and so it was expected that similar connections would be identified here. However, Study III was based on a small sample of only 17 children. To that should be added that data were missing for four children from the age of 3 years (timepoint 1), meaning that the sample at that timepoint was even smaller. This could clearly have exerted an effect on calculations. Additionally – as is evident in all studies included in the present thesis – the variety within the group of IA children with UCLP seems to be very great. This also risks affecting the outcomes of correlation analyses. Hence all correlations reported in the present studies must be interpreted with caution.

In-depth studies of speech production most often include analyses of phonetic transcriptions. It is well known that the severity of a child's speech disorder increases the difficulty of the task of phonetically transcribing his or her speech (Kent et al., 1999; Shriberg & Lof, 1991). Previous studies of children with CLP have shown that it is difficult to transcribe cleft-palate speech. In particular, the transcription of passive speech characteristics has been shown to yield low inter-transcriber agreement (Brunnegård & Lohmander, 2007). This was also true here in Study I, where some of the articulation variables manifested low agreement. In addition, inter-rater agreement in that study for the overall perceptual assessment of VP competence, with median percent values, was between 76% and 84% for all three pairings of raters. This is also in line with previous studies (Brunnegård & Lohmander, 2007; Lohmander et al., 2017b) showing that inter-rater agreement on variables linked to VPI, such as hypernasality, is often low. Because of these previous reports of low agreement on hypernasality, it was decided to analyse VP competence using VPC-Sum in Study II in order to limit the effects of low agreement for hypernasality. Additionally, all phonetic transcriptions in Studies I–III had the same master transcriber, who had not been involved in any of the previous speech assessments or interventions at the hospital for any of the participant children with CLP.

The importance of using standardised methods, performing careful and detailed analyses, collecting as much data as possible in a systematic manner and carefully choosing comparison groups is even greater when the group studied is heterogeneous, as is the group of IA children. All of the above must be kept in mind when the results from each study are interpreted. The data used in the present thesis are based on detailed and standardised assessments and analyses, in combination with the systematic collection of data from medical charts and parental ratings.

The speech and language acquisition of IA children with cleft lip and palate is unique. By learning from their developmental trajectories, we may have an opportunity to gain more knowledge about the speech and language acquisition of all children with cleft lip and palate. The overall results of the analyses of speech production showed that consonant proficiency, consonant errors and VP competence improved a great deal between the age of 3 years and the age of 7–8 years. In addition, the children's consonant inventories increased significantly. However, there was great within-group variability on all variables assessed and at all ages. At group level, the speech-production development of the IA children with UCLP seems to be slow. Even at school age, many children present with low consonant proficiency and a considerable amount of consonant errors – both errors related to the cleft palate and developmental errors usually seen only in younger children. Hence it is evident that the speech development of the IA children with UCLP is slower, but what is alarming is that the gap to their same-age peers remains wide at school age.

The IA children with UCLP in this thesis did not differ on measures of expressive and receptive language from IA children without cleft lip and palate. However, the finding that all IA children (at group level) scored low on measures of expressive language gives cause for concern; there is a need for future studies of school-aged IA children.

Conclusions and clinical implications

- Speech production – i.e. consonant proficiency, consonant inventory and perceived velopharyngeal competence – improved a great deal with age in the IA children, but there was much variety among them. In fact, most of the IA children had low scores on PCC and 20% of them still had an incompetent velopharyngeal competence at school age.

- Consonant errors decreased by age, but even at the age of 7–8 years, many children still had oral retracted articulation and developmental speech characteristics.

- Consonant proficiency and consonant inventory at the ages of 3 years, 5 years and 7–8 years were not related to age at adoption, suggesting that the age at adoption for the IA children with unilateral cleft lip and palate (UCLP) in the present thesis had minor effects on speech outcome.

- Only weak correlations were found between age at adoption/exposure time and measures of language ability, but there was a tendency that an earlier age at adoption yielded greater sentence length at the age of 7–8 years.

- There were no differences between IA children with and without UCLP on measures of language ability, indicating that the cleft lip and palate itself did not affect language ability in the IA children with UCLP.

- Expressive language, measured using the Recalling Sentences subtest and through the retelling of a story, was difficult for many school-age IA children, irrespectively of whether they had UCLP or not, suggesting that expressive language needs to be focused upon in language assessments of IA children.

- Most parents of IA children with UCLP reported concerns regarding their child's speech ability when he or she was 7–8 years old.

- Since a high proportion of children with UCLP showed expressive language difficulties and since there were children with poor receptive language skills, it is important to assess overall language performance.

- There is a need to focus on language development in children who have been internationally adopted, regardless of whether they have cleft lip and palate or not.

The present thesis found that many previous studies of internationally adopted children with cleft lip and palate were based on limited data and in some cases used questionable methods. One would hope that future studies make use of the multi-professional teams usually collaborating to help children with CLP. Those teams are very important for increasing overall knowledge about children with cleft lip and palate and for increasing the quality of research. This thesis was made possible thanks to a multi-professional team and its high-quality standards when it comes to systematically collecting data, performing standardised assessments and welcoming people from other clinical areas to analyse their data. While it may not have been mandatory yesterday to carry out research based on international recommendations relating to the standardised collection of data, to assessments and to analyses, it definitely should be today.

Many children with UCLP in this thesis still manifested developmental speech characteristics at the age of 5 years, and some of them continued to exhibit such consonant errors even at the age of 7–8 years. Since developmental speech characteristics are not thought to be related to the cleft palate, it is important to target them early on in the planning and execution of speech therapy.

The present thesis also found a high proportion of children with poor expressive language. There is a need for more knowledge about the language development of all IA children. In fact, all IA children who are referred to an SLP, regardless of the reason for the referral, should be offered an overall speech and language assessment enabling a comprehensive assessment of all language areas. The language ability of IA children needs to be placed in focus if, for some reason, they attract special attention at school because of a failure to learn, to reach curricular goals or to follow school routines. Additionally, it is crucial to provide parents, especially parents of IA children, with information about speech and language development and advice on how to expand and develop a child's different language abilities. Further, the child-healthcare services also need more knowledge about the children who are adopted nowadays so that they can better identify those children who may need further interventions in speech, language and communication areas. It is important that all healthcare professionals who see adopted children should pay attention to those children's potential need for such interventions.

Parents of IA children with CLP need early information about how to stimulate not only their child's speech ability as such but also his or her language development in general. There is a risk that the fact that a child has a CLP and undergoes the attendant assessments and interventions may overshadow the importance of stimulating the child's language development as well. Early interventions and advice on vocabulary, syntax and narratives may help the child develop even more in those areas of language. However, because of the inadequacy of the evidence base available within that area today, there is a need for future studies regarding early language interventions targeting this group of children.

Finally, it has become evident during work on the present thesis that there is a need to develop, translate and validate additional standardised language tests in Sweden. Most Swedish SLPs are working in the area of child language today, and in order for them to make more reliable assessments, they need access to additional standardised test materials.

Future Perspective

The samples of children studied in the present thesis represent a small group of children, and international adoptions of children with CLP have declined drastically. However, according to the national Swedish adoption organisations, almost all children adopted nowadays have some sort of special need. We cannot know whether there will be another change in the world of international adoptions and in the types of diagnosis that future adopted children may have. Something akin to the sudden change in 2008, with a very large number of internationally adopted children with CLP, may well happen in the future.

We hope that the results relating to speech production obtained in the present thesis will have clinical implications and will increase the knowledge of professionals working with all children who, for some reason, do not receive an early palatal closure. Some children in developing countries, where opportunities to receive early multiprofessional interventions are small, are offered treatment by NGOs such as Operation Smile. We hope that our speech-production results may provide some guidance to the professionals working within these organisations, perhaps making them better able to inform parents about potential outcomes.

Children with special needs will continue to be adopted to Sweden and Europe. We believe that it would be of importance to follow up the specific groups of IA children with and without UCLP in the future. During the time spent working on the studies in the present thesis, a great many questions have emerged and hypotheses been put forward. Is intelligibility affected in IA children with UCLP since they have so low speech-production scores? What is the effect of the low PCC scores in combination with a marginally incompetent VP function, and does this affect intelligibility even more? Does a low speech-production score affect social communication and relations with peers? What about the children's own opinions about their speech, language and communication? What about academic performance in all IA children? What types of speech and language interventions are the most effective when it comes to enhancing speech and language development in children with speech impairments and poor expressive language skills? There are many questions still in need of an answer and many ideas for future studies. During the time of data collection, almost all parents of the IA children generously told their child's stories about daily life and previous journeys in life. It would be a privilege to meet these children again and ask whether they would like to tell their own stories about having a CLP and being adopted, and to perform a qualitative study on the basis of those stories.

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