# Oral Motor Function, Voice, Speech and Language in Children with Tonsillar Hypertrophy in Relation to Surgical Outcome

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# List of original publications

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

- I Lundeborg, I., McAllister, A., Graf, J., Ericsson, E. & Hultcrantz, E. (2009) Oral motor dysfunction in children with adenotonsillar hypertrophy effects of surgery *Logopedics, Phoniatrics Vocology*, 34:3, 111-116
- II Lundeborg, I., Ericsson, E., Hultcrantz, E. & McAllister, A.

  Acoustic and perceptual aspects of vocal function in children with adenotonsillar hypertrophy -effects surgery. Submitted for publication.
- III Lundeborg, I., Ericsson, E., Hultcrantz, E. & McAllister, A.
  Influence of adenotonsillar hypertrophy on s-articulation in children Effects of surgery. Accepted for publication in *Logopedics*, *Phoniatrics*, *Vocology*.
- IV Lundeborg, I., McAllister, A., Samuelsson, C., Ericsson, E. & Hultcrantz, E. (2009) Phonological development in children with obstructive sleep-disordered breathing *Clinical Linguistics and Phonetics* 23:10, 751-761.

## **Abstract**

The aim of this thesis was two-fold: first, to evaluate four different functional aspects of the speech and language spectrum; oral-motor function, voice, /s/-articulation and phonology in preschool children with tonsillar hypertrophy before and after surgical treatment. The second aim was to investigate weather the outcome of surgery was equal for two surgical techniques; tonsillectomy or tonsillotomy combined with adenoidectomy when necessary. In all included publications (I-IV), 67 children on waiting list for tonsil surgery and randomized to either tonsillectomy (33) or tonsillotomy (34) participated. The children were assessed and audio-recorded within a month before surgery and six months postoperatively. Results were compared to age-matched control groups.

In the first study, oral motor function was assessed using the Nordic Orofacial Test-Screening, NOT-S, consisting of a structured interview and a clinical examination. Before surgery, the children in the study group differed in all domains of the structured interview in comparison to age-matched controls and in the clinical examination regarding the parameters "deviant lip position" and "trouble nose-breathing". Postoperatively oral motor functions were normalized in both surgical groups and no differences to age matched controls were observed.

In study two, recordings of three sustained vowels (/a, u, i/) and 14 words elicited by picture naming were analysed both perceptually and acoustically. Compared to the controls, significant differences were found in the study group preoperatively with higher ratings on Visual Analogue Scales (VAS) for the voice quality parameters "hyponasality" and "compressed/throaty" and also lower for "pitch". Significantly higher values on all studied perturbation measures (jitter, shimmer and Noise to Harmonics Ratio) were found. Regarding center frequencies of formants, the study groups had lower F3 values for /u / and also lower F2 and F3 for / i / compared to age-matched controls. After surgery there were no significant differences between the perceptual ratings of voice quality of the two surgical groups and the corresponding controls. The acoustic analyses showed a decrease in all the measures of perturbation for the study group after surgery with a slight difference between the two surgical groups. The children in the tonsillotomy group had higher shimmer value for /u/ and

higher NHR for / $\alpha$ /. In comparison to the older controls significantly higher values were found an all perturbation measures and the difference seen regarding formant frequencies for the /i/-sound in comparison to controls still remained. The significantly lower third formant (F3) of the /u/-sound also remained. When comparing pre- versus postoperative results for the surgical group as a whole, a decrease was found on all perturbation measures postoperatively, however the differences were not statistically significant. A significant increase was found in formant 3 for / $\alpha$ / and / $\nu$ /.

The material used in the third study were speech samples containing the /s/-sound and elicited by picture naming and sentence repetition. Before surgery the study group was rated to have more indistinct /s/-sounds than age-matched controls. The acoustic analyses showed that the study group had lower spectral peak values for the /s/-sound than controls. After surgery the operated children's /s/-production did not differ perceptually from the older controls, neither as a whole group nor when divided according to surgical methods. Regarding the acoustic analyses however, the study groups differed from the age-matched control group showing that noise duration was longer and the peak location higher in the study groups.

In study four, a Swedish phonology test was performed and transcribed phonetically. The transcription of each child was analyzed in terms of phonological processes and categorized into one of six developmental stages according to the model developed by Nettelbladt (1983) and adapted by Sahlén, Reuterskiold-Wagner, Nettelbladt & Radeborg (1999). A majority of the children in the study group (62.7 %) showed a slowed phonological development preoperatively (developmental stages 0-4), compared to the age-matched control group. Postoperatively the children in both surgical groups had improved their phonological skills. However, they were still behind in comparison to age-matched controls and the difference was even larger than before surgery.

The results of this thesis project have clinical relevance for both speech and language pathologists (SLP's) and ear-nose and throat-surgeons (ENT-surgeons). SLP's must be aware of the potential impact of tonsillar hypertrophy on oral-motor function and the speech and language spectrum to be able to help affected children adequately and ENT- surgeons should include oral motor and speech and language problems as additional indications for tonsillar surgery.

# **Abbrevations**

A.M. Anita McAllister

C.S. Christina Samuelsson ENT Ear, Nose and Throat

f0 Fundamental frequency

F1 First formantF2 Second formantF3 Third formant

GRBAS Grade, Roughness, Breathiness, Asthenia, Strain Scale

Hz Herz

I.L. Inger Lundeborg

LI Language Impairment

LTAS Long Time Average Spectra

NHR Noise to Harmonic Ratio

ND Noise Duration

NOT-S Nordic Orofacial Test-Screening

OME Otitis Media with Effusion

OSDB Obstructive Sleep-Disordered Breathing

PL Peak Location
QOL Quality of life

SLP Speech and Language Pathologist

SVEA The Stockholm Voice Evaluation Approach

TE Tonsillectomy
TT Tonsillotomy

VAS Visual Analogue Scales

## Introduction

Four different but related functional aspects of speech are investigated in this thesis: oral motor ability, voice, articulation and language in children with tonsillar hypertrophy.

Problems with these functions have great impact on general health and well-being (Casselbrant, 1999; Trulsson & Klingberg, 2003).

Structural abnormalities in the vocal tract are known to affect several orofacial functions (Chapman, 1993; Pamplona, Ysunza, Gonzalez, Ramirez, & Patino, 2000; Valera et al., 2003) and when hampered, this not only affects the child, but often also the whole family (Evans Morris & Dunn Klein, 2000; Trulsson & Klingberg, 2003). One such structural abnormality in the vocal tract is adenotonsillar hypertrophy. A relative hypertrophy of the adenoid and the tonsils is common in children between the ages of 3 and 5 years as a result of a developing immune system. In many children the enlargement and the subsequent decrease of oropharyngeal airspace causes obstructive symptoms of varying degree and different functional aspects are affected (Valera et al, 2003; Anand, Vilela, & Guarisco, 2005; Mora, Crippa, Dellepiane, & Jankowska, 2007). The children with obstructive problems are usually treated with adenoidectomy and tonsil surgery, which is the operation most frequently performed in children (Younis & Lazar, 2002). With regard to tonsil surgery, two different methods are currently used, tonsillectomy (TE) and partial tonsillotomy (TT) (Hultcrantz, Linder, & Markström, 1999; Densert et al., 2001). The questions studied in this thesis were: 1. To what extent oral motor function, articulation, voice and language are affected by tonsillar hypertrophy and 2. Whether outcome of the two surgical methods is equal with respect to the studied functions.

# **Background**

## Historical background

Until the 18<sup>th</sup> century most medical care in Sweden was conducted by autodidact barber surgeons mainly dealing with amputations and other external disorders. Children were rarely treated and 20% of all children died within their first year of life (Fåhreus, 1970). From the second half of the 18<sup>th</sup> century Swedish medicine developed rapidly and by the end of the century hospitals were opened in several cities. Simultaneously, attention was brought to paediatric care with the publication in 1771 of Nils Rosén von Rosenstein's ground-breaking textbook on paediatrics (Radbill, 1966). The first children's hospitals in Sweden opened in the mid 19<sup>th</sup> century (Ohrlander, 2004).

Speech impediments in children were primarily a concern for teachers in the beginning of the 20<sup>th</sup> century in Sweden. They often classified children with deviant speech as also being cognitively impaired (Fritzell, 2003). Physicians in Germany and Austria were the first to differentiate between children with general mental retardation and children with speech and language impairments and constituted a new medical speciality, antecedent to today's phoniatrics (Nettelbladt & Samuelsson, 1998). In Sweden, physicians Alfhild Tamm and Karl Weinberg, were inspired by their colleagues abroad and their pioneer work led to the opening of the first phoniatric clinic in Sweden 1929 and the recognition of phoniatrics as a medical speciality in 1931 (Fritzell, 2003). The first phoniatricians trained speech therapists and in 1964 the first formal speech pathology and therapy program started at Karolinska Institutet in Stockholm. Since then the field of work for speech language pathologists has grown substantially and not only covers speech and language impairments, but also oral motor function including eating and swallowing problems.

# Oral motor function/orofacial function

Orofacial function includes several vital actions such as breathing, chewing and swallowing, articulation and facial expression (Bakke, Bergendal, McAllister, Sjogreen, & Asten, 2007). All these functions have great impact on quality of life (QOL), which is a notion that encompasses both subjective perceptions of well-being and the ability to perform everyday activities and objective functioning (Gotay & Moore, 1992).

Oral motor function is gradually developed from birth onward to puberty. During the first two-three years of life there is a rapid growth of the involved structures as well as a maturation of the neural system. The orofacial structures are remodelled so that they approach an adult-like configuration. These developmental changes are not determined by growth factors alone. Form and function interact strongly and the structures are shaped by their use (Kent, 1999). After four years of age, a refinement or fine tuning of oral movement organization takes place (Sharkey & Folkins, 1985; Robbins & Klee, 1987) and variability between different children's performance decrease (Robbins & Klee, 1987). Young boys have slower maturation than girls, but seem to catch up after the age of five (Smith & Zelanik, 2004; Cheng, Murdoch, Goozee, & Scott, 2007).

Oral motor dysfunction can be very disabling and affect quality of life (QOL) negatively. QOL refers to an individual's emotional, social and physical well being, including their ability to function in the ordinary tasks of living. When health status and functional status are in focus as in studies of single disease states, patient groups or areas of function, the notion health-related quality of life (HRQL) is used (Guyatt, Feeny, & Patrick, 1993). A reduced HRQL is reported in children with breathing obstructions, such as asthma (Cohen, Noone, Munoz-Furlong, & Sicherer, 2004) and children with speech problems (Karande, Bhosrekar, Kulkarni, & Thakker, 2009).

The primary role of the oral motor system is that of digestion and breathing, but it also plays a significant role in speech production.

### Voice

The voice is an integral part of human speech and originates in the larynx. A controlled exhalation sets the vocal folds in vibration, the aerodynamic energy is converted to acoustic energy and sound is produced. This acoustic energy is distributed to the oral cavity where it is shaped into different speech sounds. Thus both laryngeal and supralaryngeal features contribute to voice quality. A resonant voice has been described as an ideal mix of vocal fold vibrations and ample reinforcement by the vocal tract (Titze, 2001). Voice is present when producing all vowels and many of the consonants. In addition to its role in speech production, voice is also important in the expression of emotions and in singing (Colton, Casper, & Leonard, 2006). In a broader sense, voice quality can be seen as the auditory perception of an individual speaker's voice. Perceptually, voice quality is a cumulative abstraction over a period of time of a speaker-characterizing quality (Laver, 1980).

The human voice develops from the new-born infant's cry and undergoes an increasingly complex differentiation towards different acceptable vocal behaviours in different situations. This development is largely dependant on the growth of and the motor control of the larynx but also on cognitive and emotional maturation (Colton et al., 2006). Since the characteristics of voice function are multidimensional, adequate assessments of voice must include both objective and subjective measures. In view of the fact that voice quality is perceptual by nature, perceptual characteristics have greater intuitive meaning than many instrumental measures (Oates, 2009). In clinical practice perceptual evaluation is most commonly used. However, clinical applications of perceptual-auditory judgements are based on the assumption that listeners have a common understanding of the perceptual labels of voice and use the same scale value to assess a given voice sample (Kent, 1996). With adequate control of factors that are known to affect auditory-perceptual judgements such as type and complexity of the rating task, listener's background and experience, perceptual evaluation can be fairly stable (Oates, 2009). Several studies have created systems to analyze voice quality perceptually and different perceptual evaluation protocols have been developed. Most of them are developed for the assessment of pathological voices and the GRBAS (Grade, Roughness, Breathiness, Asthenia, Strain) scale developed by Hirano (1981) and for Swedish voices, SVEA (The Stockholm Voice Evaluation Approach) (Hammarberg & Gauffin, 1995; Hammarberg, 2000) are often referred to in the literature (Bele, 2005). Since it can be assumed that vocal characteristics are perceived along a continuum rather than stepwise, the use of continuous scales such as the visual analogue scale (VAS) offers more detailed information (Sederholm, McAllister, Sundberg, & Dahlqvist, 1993).

Different types of voice stimuli are used in evaluations of voice quality, in both running speech and sustained vowels. Both types have advantages and disadvantages. Running speech provides a more natural speech but in a long sentence, opposing features can be heard in different sentence parts. The voice can, for example, be hypofunctional at the beginning of a sentence and hyperfunctional at the end (Bele, 2005). Therefore the recommendation is to use a combination of both sustained vowels and connected speech. To assess voice quality with good reliability is a difficult task and requires professional experience (Bassich & Ludlow, 1986). This difficulty is well known and several researchers recommend that perceptual assessments are complemented by acoustic analyses. The acoustic signal bridges the acts of speech production and speech perception. Therefore, acoustic analysis is informative about both a talker's behaviour and a listener's perception of the signal generated (Kent & Kim, 2008).

If, for some reason, the vocal apparatus is subjected to a particular long-term muscular adjustment such as a protrusion of the tongue or a structural interference with the settings of the velopharyngeal muscles due to tonsillar hypertrophy (Henningsson & Isberg, 1988) the auditory perception of the voice is often affected. Contributing factors to these changes are resonatory phenomena occurring above the vocal folds such as nasality or a compressed/throaty voice quality.

## Articulation

Speech is the acoustic output of movements in the vocal apparatus intended to express thoughts, feelings and expressions of the speaker. It is one of the most complex motor behaviours that humans routinely master and requires spatial and temporal coordination of more than 70 muscles ranging from the abdomen through the whole vocal apparatus to the lips (Cheng et al., 2007). The adult mastery of fluent articulation takes many years to develop and the developmental journey from prelinguistic vocalizations to mature speech sound production takes place starting shortly after birth and ends in late adolescence. The course of this development has been studied by many different researchers throughout the world and the order of acquisition is fairly consistent across different languages.

Speech involves the respiratory system including the larynx and the upper airway system. Articulation refers mainly to the movements of the structures in the upper airway systems that result in the acoustic characteristics of the speech sounds.

Children's earliest articulatory productions consist of roughly specified gestures that are not organized in the precisely timed manner that characterizes adult articulation. Normal babbling manifests the infant's growing capacity to produce sounds that resemble speech and can be regarded as the foundation for speech (Oller, 2000). Two processes are involved in the following development towards mature speech; differentiation and tuning of individual articulatory gestures, and improvement of coordination among gestures across an utterance (Browman & Goldstein, 1989). Two steps are considered important. The first is the emergence of canonical babbling, around seven months of age, with strings of two contrasting oral gestures, the narrow or complete constriction of a consonant and a wider constriction associated with a vowel (Studdert-Kennedy & Goodell, 1995). Regardless of native language, complete closures and openings associated with stop consonants and the open vowel /a/ are

common in children's first sound productions. The perception of these segments may have more to do with the perceiver than the child (Nittrouer, 1995). At the beginning of canonical babbling the sequences are often produced without actual reference and more as a pure motor exercise. The next step is differentiation of the syllable into independent gestural components where the consonant and vowel-like sounds differ in successive syllables. This richer and more variegated babbling is interwoven with the canonical babbling stage and the development of meaningful sound sequences which seem to arise independently of adult language (Stackhouse & Wells, 1997). The subsequent stage is the onset of words that emanate from adult form, referred as the onset of real speech. Real speech begins with indistinct productions that must be deciphered with sophistication and shaped by the caretaker towards some potential meaning (Oller, 2000). By the age of 12 months most children enter the phase of real speech with the emergence of first words. The first word forms are determined by the child's articulatory capacity and consist of a single undifferentiated gestalt of gestures (Stackhouse & Wells, 1997). Now the input aspect of speech is added to the articulation in that children produce words that they have heard and understood. In the next step, during the child's second year, the range of phonetic forms in the output increases rapidly. Subsequently the child's articulation is gradually getting less variable and the correspondence with adult forms is becoming more systematic. By the age of four a majority of Swedish children are able to articulate all speech sounds, but before the age of six years the sound production is often variable (Nettelbladt & Salameh, 2007). If the timing, place and manner of articulatory movements are incorrect (including appropriate speed and force), the acoustic output is compromised. One of the most difficult aspects of articulation is the production of the /s/-sound. (Niemi, Laaksonen, Ojala, Aaltonen & Happonen, 2006). It demands a great degree of articulatory precision and a variation of one millimetre in the position or shape of the articulators results in a great difference in acoustic output (Ladefoged & Maddieson, 1996). There can be different underlying reasons for a child's inaccurate production. Apart from an abnormal structure or function in the articulators, it may reflect immaturity or abnormality in the acquisition of the sound system of the child's native language (Norbury, Tomblin, & Bishop, 2008).

# Language and language development

To paraphrase the famous quotation "Language is the dress of thought" by Dr Samuel Johnson (Johnson, 1781) you could say that speech is the dress of language. Without a communicative intent intermediated by language symbols, speech has no meaning.

Language formulation is composed by word retrieval, mapping of the phonology of these words and grammatical framing. It is a mental process that precedes the planning, programming and execution of speech movements. Language as a system is said to be organized in different levels, with different units of analysis. These levels are phonology, grammar, semantics and pragmatics (Mogford & Bishop, 1993).

Language acquisition involves a range of smoothly integrated cognitive and language processes. There is a close mutual relation between thought and language in the child's development and through this, thought becomes verbal and speech becomes rational. Essential prerequisites in this development are experience and social interaction (Vygotsky, 1986). There is also a close relationship between the developmental pathways of the different organizational levels of language. In several studies, the importance of the development of phonological ability for the other domains of language has been established (Menyuk & Looney, 1972; Cassidy & Kelly, 2001; Fitneva, Christiansen, & Monaghan, 2009).

The importance of the phonological development has been the primary motivation for the focus on the phonological domain in his thesis. Phonology refers to a child's ability to use speech sounds according to the adult norm of the specific language to convey meaning (Stackhouse & Wells, 1997). The process of phonological acquisition in children has been described as a process of organizing and learning the mappings between the auditory speech input and the gestural control of speech output (Snowling & Hulme, 1994). The child's phonological development goes from a stage where there is no specification in terms of sound segments and their respective distinctive phonological features, to an organization where vowels and consonants are specified segments with distinctive phonological features (Samuelsson, 2004). Phonological ability also improves through increased capacity to produce adult sounds and combine them into more complex phonological structures (Ingram, 1979).

By the age of five, typically developed children master both the fundamental structural aspects of language and the ability to determine how to use both verbal and non-verbal language to understand and convey a wide variety of messages according to context

(pragmatic knowledge) (Baird, 2008). However, there are children who do not develop their language abilities according to the expectations of the caretakers. The causation is obvious in many cases, but children with language problems without an identifiable ethiology form a relatively large and heterogeneous group (Gibbon, 1999). One of the major debates within the study of developmental language disorders concerns whether children's impaired language is caused by incomplete linguistic knowledge or processing limitations (Marinis, 2008). Another issue is to what extent a structural abnormality affects the developmental course. There is evidence indicating that children with cleft lip and palate have deficits in the phonological domain of language. These findings are interpreted as the result of phonetic compensations that have been integrated in their phonological system and thus, become part of their language (Chapman, 1993; Pamplona et al., 2000).

## Adenotonsillar hypertrophy

The palatine tonsils, located laterally at the entrance of the pharynx, the adenoid superiorly and the lingual tonsil at the tongue base together form a ring of lymphoid tissue, called Waldeyer's ring. The lymphoid tissues of Waldeyer's ring are thought to play an important role in host- defence against upper respiratory infections (van Kempen, Rijkers, & van Cauwenberge, 2000).

During the first four years of life, the respiratory tract is exposed to a multitude of infections. In response to these infections, most children between the ages of 3 to 5 years have a relative hypertrophy of the lymphoid tissue of the upper part of the pharynx. During these years, the size of the oro-naso-pharyngeal space is not yet fully developed. Thus a prominent Waldeyer's ring occupies a relative large part of the space in the upper airway and causes functional problems for many children (Casselbrant, 1999; Darrow & Siemens, 2002).

# Tonsillar hypertrophy and different functional aspects

Tonsillar hypertrophy is associated with a wide range of problems. The decreased oropharyngeal airspace causes obstructive breathing problems of different degree. The symptoms increase when the child is lying down and the tonsils fall backwards and may meet in the midline causing sleep disturbances ranging from simple snoring to obstructive sleep apnoea (Behlfelt, Linder-Aronsson, McWilliam, Neander, & Laage-Hellman, 1990). The diminished lumen forces the child to increase the respiratory effort to maintain an adequate airflow. The approximation of the tonsils give rise to the Bernoulli effect creating a negative pressure in the oropharyngeal airspace which causes the compliant airway structure to collapse and may result in stoppage of the air flow (Rahbar, 2004). In Obstructive Sleep-Disordered Breathing (OSDB) the children have night-time, mouth-breathing, pauses in breathing, snoring, gasping, sweating, restless sleep and in many cases enuresis (Darrow & Siemens, 2002; Benninger & Walner, 2007).

Daytime functioning is also often affected by the pharyngeal obstruction. Affected areas are breathing, chewing and swallowing, speech and voice and also cognitive functions. The reduced airspace results in oral breathing, which, in turn, causes postural alterations of several orofacial structures such as a habitual open mouth, lower-anterior position of the tongue and a lower position of the hyoid bone. In the long run, these postural alterations may also have effects on dental occlusion and craniofacial development (Hultcrantz et al., 1991; Lofstrand-Tidestrom, Thilander, Ahlqvist-Rastad, Jakobsson, & Hultcrantz, 1999; Lofstrand-Tidestrom & Hultcrantz, 2007). The impact on chewing and swallowing results in difficulties swallowing solid food. A failure to thrive, which is associated with dysphagia, is reported in connection with tonsillar hypertrophy (Kara, Ergin, Kocak, Kilic, & Yurdakul, 2002: Ericsson, Lundeborg, & Hultcrantz, 2009).

Several aspects of the speech spectrum such as voice characteristics and articulation are reported negatively affected by tonsillar hypertrophy (Valera et al., 2003; Maryn, Van Lierde, De Bodt, & Van Cauwenberge, 2004; Mora et al., 2007). Regarding voice quality there are reports of hypernasality as a result of the enlarged tonsils hampering velopharyngeal closure (Henningsson & Isberg, 1988). Others state that hyponasal speech is common, especially when both the tonsils and the adenoid are enlarged (Rahbar, 2004). Some characterize the impact on resonance by cul-de-sac resonance with a hollow and muffled sounding voice (Shprintzen, Sher, & Croft, 1987; Wetmore, Muntz, & McGill, 2000). Problems with articulation of dento-alveolar consonants and voice resonance have been previously reported (Ahlqvist-Rastad, Hultcrantz, & Svanholm, 1988; Maryn et al., 2004; Mora et al., 2007; Salami, Jankowska, Dellepiane, Crippa, & Mora, 2008). Obstructive sleep disordered breathing (OSDB) is also reported to be a co-morbid factor for substantial behavioural, neurological and cognitive deficits such as hyperactivity, mood disturbances, reduced memory and attention, as well as impaired academic results (Chervin, Ruzicka, Archbold, & Dillon, 2005; Kheirandish & Gozal, 2006).

Children with tonsillar hypertrophy often have a reduced quality of life with emotional distress, reduced daytime functioning and a high degree of concern in care givers apart from the sleep disturbance compared to age matched controls (Ericsson, Lundeborg & Hultcrantz, 2009).

## Tonsillar surgery

## Indications for surgery

Indications for tonsillar surgery have changed over the years. Until the 1960s, when the use of peroral antibiotics for children was developed, tonsil surgery usually was performed to avoid complications of scarlet fever and recurrent tonsillitis (van den Akker et al., 2003). From the 1960s onwards the number of tonsil operations decreased and the indications for surgery changed. In the past 15-20 years, the two most important indications for surgery have been obstructive breathing and frequent episodes of tonsillitis (van den Akker et al., 2003; Ericsson, Hemlin et al., 2009).

## Surgical methods

Tonsil surgery is the operation most frequently performed in children (Younis & Lazar, 2002; Koempel, Solares, & Koltai, 2006). In Sweden the prevalence is about 10.000 operations annually (Ericsson, Hemlin et al., 2009). Surgical techniques and instrumentation have been refined over the years and surgery can be done either "cold" with blunt dissection or aided by different electrically powered scalpels or radio-frequency instruments (Ericsson, 2007). The most common surgical technique is tonsillectomy (TE) with or without adenoidectomy. In standard tonsillectomy the entire tonsil including the capsule is removed. Tonsillectomy is associated with considerable postoperative pain and significant morbidity during the recovery period (Hultcrantz et al., 1999; Anand et al., 2005). There is also a certain risk, of complications in the form of haemorrhage, estimated to 1-5 % in children (Windfuhr & Chen, 2002).

An alternative surgical method is tonsillotomy also known as partial tonsillectomy. In this method only the obstructive tissue is removed leaving a normal sized tonsil within the tonsillar pouch (Hultcrantz, Linder, & Markström, 1999; Densert et al., 2001; Hultcrantz & Ericsson, 2004; Ericsson & Hultcrantz, 2007). Several studies have shown that tonsillotomy gives lower primary morbidity with less postoperative pain in comparison with tonsillectomy (Anand et al., 2005; Koempel et al., 2006; Ericsson, & Hultcrantz, 2007; Ericsson, Lundeborg & Hultcrantz, 2009). The risk of secondary postoperative bleeding (> 24 hours after surgery) is 0,1-1 % for tonsillotomy compared to 3-11 % for tonsillectomy (Koltai, Solares, Mascha & Xu, 2002; Solares, Koempel, Hirose, Abelson, Reilly, Cook, April, Ward, Bent III, Xu & Koltai, 2005; Ericsson & Hultcrantz, 2007; Ericsson, Lundeborg & Hultcrantz, 2009, Gan, Tomlinson & El-Hakim, 2009).

## **Aims**

The overall aim of this thesis's project is to evaluate four different functions in preschool children with symptoms of tonsillar hypertrophy before and after two types of tonsil surgery, in comparison to age-matched healthy controls. Evaluated areas are oral motor function, voice, articulation and language development. The two surgical methods are tonsillectomy or tonsillotomy combined with adenoidectomy when necessary. The purpose is to compare surgical outcome with respect to mentioned functions.

## Specific aims

- To evaluate oral motor function in preschool children before and after planned surgery due to obstructive breathing problems in comparison to age-matched healthy controls (Study I).
- To evaluate voice function perceptually and acoustically in preschool children before
  and after planned surgery due to obstructive breathing problems in comparison to agematched healthy controls (Study II).
- To investigate whether /s/-articulation is affected in preschool children before and after planned surgery due to obstructive breathing problems in comparison to agematched healthy controls (Study III)
- To investigate whether phonological development is affected in children with tonsillar hypertrophy and obstructive sleep disordered breathing and to study the outcome of two types of surgery in comparison to age-matched healthy controls (Study IV).

## **Methods**

## Study design and participants

This thesis' project is a prospective randomized controlled trial approved by the Human Research Ethics Committee in Linköping (No 03-448 and No M138-08). The thesis includes four separate studies.

#### Inclusion and exclusion criteria

Children eligible for the project were, 4;5-5;5 years old, with tonsillar hypertrophy and sleep disordered breathing with or without recurrent tonsillitis. These children were initially randomized from the ordinary waiting list for tonsil surgery at three ENT-clinics in the southeast region in Sweden, a region that, in socioeconomic terms, could be described as in the mid level. The criteria for exclusion were treatment with antibiotics for upper air-way infections during the last three months, previous treatment for peritonsillitis, records stating small tonsils, complex special needs (children with cognitive deficits and/or physical disabilities) and children with non-Swedish speaking parents.

## Randomization procedure

The randomization was carried out according to the method of Zelen (Zelen, 1981). One hundred and eighteen consecutive children on the waiting lists for tonsil surgery were randomized to either tonsillectomy (TE) or tonsillotomy (TT). The procedure was done using a randomly computer-generated sequentially numbered list. An independent person drew from this list and assigned odd numbers to tonsillectomy and even numbers to tonsillotomy. After this the children's caretakers received written information about the study and the type of surgery their child would receive and were invited to participate. Four children dropped out due to randomization errors (three TE and one TT). Thirty-six families declined participation (22 TE and 14 TT) at an early stage. Ten children were excluded according to the exclusion criteria. The caretakers of one child declined surgery (TE) just before it was due and one child (TE) had recovered spontaneously before surgery. Participants and withdrawals are illustrated in figure 1. A total of 67 children (33 TE and 34 TT), aged 50 to 65 months, and consisting of 28 girls and 39 boys were included in the project. Six of these children had undergone an adenoidectomy earlier. None of the participating children had any speech or language therapy prior to the study. Six of the children had otosalpingitis at the preoperative examination with

need for grommet insertion. All caretakers signed informed consent forms before inclusion in the studies. A total of 200 age-matched healthy and typically developed pre-school children from the same area accepted to be controls (with parental consent).

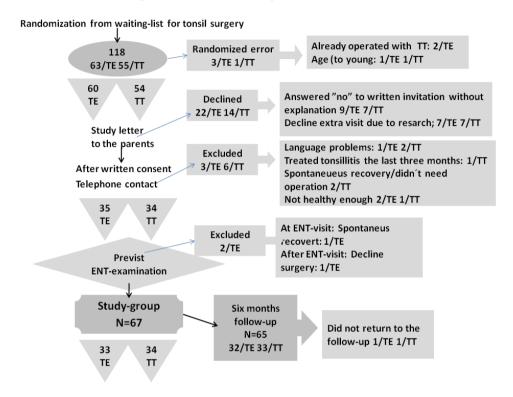


Figure 1. Participants and withdrawals

## Samples

Participants and withdrawals are illustrated in figure 1. The age and gender distribution of the children in the study group and in the different control groups are shown in table 1.

#### Study I:

Sixty-seven children attended the preoperative assessment and 33 of them received tonsillectomy and 34 tonsillotomy and 65 (32 TE and 33 TT) attended the postoperative assessment. Seventy-nine healthy age-matched controls, selected from a cohort of pre-school children were divided into two groups: 47 children for comparisons before surgery, and 32 after.

#### Study II:

The same study population as in study one participated. In this study 57 healthy controls without a history of tonsillar problems or snoring, were recorded and divided into two groups: 28 for comparisons before surgery, and 29 after.

#### Study III:

The same study population as in study one participated. Seventy pre-school children, from the same area as the surgical groups were selected to be controls to the study-groups. Inclusion criteria for controls were no history of tonsillar problems, no recurrent ear infections and no speech and language pathology contacts. Thirty-five age matched children served as controls for comparisons before surgery and 35 after surgery.

### Study IV:

Sixty-seven children and 79 age matched healthy controls (the same children as in study one) were assessed except one of the operated children who did not complete the phonology test at the postoperative assessment.

**Table 1.** The age and gender distribution of the children in the study group and in the age-matched control groups

Randomized for tonsillectomy tonsillotomy		Controls, study I		Controls, study II		Controls, study III		Controls, study IV			
Preop (n=33)	Postop (n=32)	Preop (n=34)	Postop (n=33)	Younger (n=47)	Older (n=32)	Younger (n=28)	Older (n=29)	Younger (n=35)	Older (n=35)	Younger (n=47)	Older (n=32)
ୁ11 ∂22	ୁ11 ∂21	♀ <b>17</b> ♂ <b>17</b>	ୁ 17 ∂16	ୁ21 ∂26	ୁ16 ∂16	ୁ 13 ♂15	ୁ14 ∂15	ୁ14 ∂21	ୁ19 ∂16	ୁ21 ∂26	ୁ16 ∂15
4;10 y	5;6 y	4;9 y	5;5 y	4;9 y	5;6 y	4;9 y	5;5 y	4;9 y	5;8 y	4;9 y	5;6

## **Procedure**

#### Assessments

Different methods to assess oral motor function, voice, articulation and phonology were used in this project, see below. The children with tonsillar hypertrophy were assessed within a month before surgery and six months postoperatively. The age-matched controls were tested only once. The test sessions were audio-recorded using a Marantz PMD 660 Professional Recorder and an Audiotechnica mb microphone. Formal hearing tests were not included.

## Surgery

All children received their surgery in accordance with the randomization. Thirty-three children received a tonsillectomy (TE) by cold knife and blunt dissection and 34 received a tonsillotomy (TT) by high frequency radiosurgery (Ellman 4.0 Mhz Surgiton Dual Radiowave Unit, Ellman International, Oceanside, NY). The methods have been described in detail earlier by Hultcrantz and Ericsson (Hultcrantz & Ericsson, 2004). None of the children in the tonsillotomy group experienced any complications. Two children in the tonsillectomy group had secondary bleedings and were hospitalized without need of further surgery. Adenoidectomy was performed in 25/33 children in the tonsillectomy group and 28/34 children in the tonsillotomy group during the same surgery session. Six children with otitis media with effusion (OME), three in each group also had grommet insertions.

### Assessment methods

#### Study I:

The Nordic Orofacial Test-Screening, NOT-S (Bakke et al., 2007) was used for the assessment of orofacial function in this study. NOT-S consists of two parts: a structured interview and a clinical examination consisting of six domains, see table 2. The NOT-S was adjusted for this investigation in the domains "Dryness of the mouth", "Chewing and swallowing" and "Speech". Three follow-up questions were included related to "chewing and swallowing" and dryness of the mouth. The questions were: "Is your child avoiding any food-texture?", "Is he/she chewing longer than normal?" and "Do you feel that your child is drinking a lot to be able to swallow?"

The speech items were excluded. Two principal investigators (I.L. and A.M.) performed the tests of the two study-groups. The controls were assessed as part of a master's thesis project in speech and language pathology.

**Table 2.** The Nordic Orofacial Test-Screening (NOT-S). Items included in the structured interview and the clinical examination

Structured interview	Clinical examination
Sensory function	Face at rest
Breathing	Nose breathing
Habits	Facial expression
Chewing and swallowing	Masticatory muscles and jaw function
Drooling	Oral motor function
Dryness of the mouth	Speech

#### Study II:

The children in the study group and the age matched controls, were recorded while producing three sustained vowels (/a, u, i/) and 14 words, elicited by picture naming. The criteria for the words chosen were that they should be well-known to most children and almost exclusively contain sonorants. The words were: <code>blommor /flowers/, gungar /swings/, dörr /door/, halv /half/, ballonger /balloons/, banan /banana/, räv/fox/, lejon /lion/, näbb /beak/, rädd /afraid/, hår /hair/, öra /ear/, ögon /eyes/ nalle /teddy bear/. Two principal investigators (I.L. and A.M.) did the recordings of the study group and the control group was recorded as part of a master's thesis project in speech and language pathology.</code>

#### **Study III:**

The speech samples used in this study were elicited by picture naming and sentence repetition. In some cases the word associated with the picture was elicited by repetition. The criteria for the words chosen were that they should be well-known to most children and include /s/ in conjunction with a high vowel. The sentence was composed so that the /s/sound occurred in word initial as well as in medial and final position ("Sissi och Lasse sover i sitt hus" /"Sissi and Lasse are sleeping in their house"). The children were allowed to make as many trials as needed to get the whole sentence recorded. Two principal investigators (I.L.

and A.M.) did the recordings of the study group and the control group was recorded by the author of this thesis (I.L.) and two speech and language pathology students.

#### **Study IV:**

In this study, a Swedish phonology test (Hellqvist, 1991) was performed. Two principal investigators (I.L. and A.M.) performed the tests of the two study-groups. The control material was collected as part of a master's thesis project in speech and language pathology.

## Analyses, scoring and statistics

### Study I:

Prevalence of symptoms for each item in each domain was recorded. Regarding statistics, data were expressed with descriptive statistics for demographics, and the prevalence of affected items in NOT-S. Differences between the groups with respect to non-parametric data were analyzed with the Mann-Whitney *U*-test. Changes in oral-motor function before and after surgery within the study groups were analyzed using the Wilcoxon signed-rank test.

#### Study II:

The material was analyzed both perceptually and acoustically. The perceptual analysis was done independently by three trained speech and language pathologists (SLP) blinded with respect to surgical method and pre- or postoperative status. The analysis was performed on all recorded material. A predetermined form with visual analogue scales (VAS) developed in a study of children's voice in relation to noise (McAllister, Granqvist, Sjolander, & Sundberg, 2009) and expanded for this investigation with parameters that potentially could be affected by tonsillar hypertrophy was used. The form included the following voice parameters: Hoarseness. breathiness, hyperfunction, roughness, hyponasality, hypernasality, compressed/throaty voice and pitch. The end-points of the VAS for all parameters except pitch were 'not at all' (0 mm) and 'a lot (100 mm). Pitch was represented by a 200-mm line with 'very low' and 'very high' marked, respectively, at the extremes and expected in the middle (Appendix 1) The Acoustic analysis was made on the sustained vowels using the Praat software (http://www.fon.hum.uva.nl/praat/ Version 5.1.31, Paul Boersma and David Weenink, Phonetic Sciences Department, University of Amsterdam). The following parameters were estimated: average fundamental frequency (f0), jitter percent, (local), shimmer (local), noise-to-harmonic ratio (NHR) and the center frequencies of formant 1-3

(F1,F2, F3). Demographic data were expressed with descriptive statistics. Differences between the groups were analyzed using the Mann-Whitney *U*-test both for perceptual ratings and acoustic data. Changes before and after surgery in the study groups were analyzed using the Wilcoxon signed-rank test.

### **Study III:**

A perceptual analysis of the /s/-sounds was made independently by three trained speech and language pathologists (SLP) blinded with respect to surgical method and pre- or postoperative or control status. The analysis was performed on a predetermined form with Visual Analogue Scales (VAS). The end-points were "A sharp and distinct /s/-sound" (0 mm) and "Very deviant and indistinct /s/-sound" (100 mm). The raters were also asked to tick whether they considered the overall /s/-production as deviant and if so, in what respect (Appendix 2).

Three /s/-sounds in three word positions, initial, medial and final, in conjunction with high vowels (in the words 'Sissi' and 'hus'), were also analyzed acoustically using the Praat software (<a href="http:www.fon.hum.uva.nlpraat">http:www.fon.hum.uva.nlpraat</a> Version 5.1.31, Paul Boersma and David Weenink, Phonetic Sciences Department, University of Amsterdam). Segmentation of the onset and offset of the target sound was conducted with inspection of the waveform and wideband spectrogram according to the method described by Jongman, Wayland and Wong (Jongman, Wayland, & Wong, 2000). The onset was defined as the point where the high frequency energy first appeared (also characterized by a rapid increase of zero-crossings) and the offset was determined by the intensity minimum prior to the onset of the vowel periodicity. The noise duration (the duration of the /s/-sound) was defined as the time from onset to offset and was calculated by use of the Praat software. Spectral peak estimation was made from a LongTimeAverage Spectra (LTAS) of the target sound defined as above.

Demographic data were expressed with descriptive statistics. Differences between the groups, including sex-differences, were analyzed using the Mann-Whitney *U*-test both for perceptual ratings and acoustic data. Changes before and after surgery within the study groups were analyzed using the Wilcoxon signed-rank test. Correlation analysis was made between the perceptual evaluations of /s/-articulation and acoustic measures using Spearman's rank correlation coefficient.

#### Study IV:

The recordings from the phonological test were transcribed phonetically by the author of this thesis (I.L.) using the transcription conventions of the International Phonetic Association

(1999) The phonological analysis was made in terms of phonological processes (Nettelbladt, 1983). A division between syntagmatic and paradigmatic processes was made in accordance with Nettelbladt (1983). Syntagmatic processes are defined as processes that change the phonotactic structure of the target word and paradigmatic processes are defined as context-free processes working on classes of segments.

Based on the assessment in terms of phonological processes, each child's phonology was categorized into one of six developmental stages according to the model developed by Nettelbladt (1983) and adapted by Sahlén et al. (Sahlen, Reuterskiold-Wagner, Nettelbladt, & Radeborg, 1999), se table 3.

**Table 3.** Phonological developmental stages (Nettelbladt, 1983; Sahlen et al., 1999).

- **0.** Pervasive syntagmatic restrictions; strongly restricted phonotactic structures, assimilations in mono- and disyllabic words, strongly reduced vowel system.
- 1. Pervasive reduction of clusters in combination with other optional syntagmatic processes such as assimilation in polysyllabic words, metathesis, systematic omissions of final or initial consonant and use of dummy-syllables.
- **2.** Pervasive reduction of more than one type of cluster. Optional assimilation and/or deletion of pretonic syllables.
- **3.** Reduction of one type of cluster, e.g. s-clusters. Correct production of all other types of clusters. Traces of other syntagmatic processes as metatheses especially in polysyllabic words. Pervasive paradigmatic consonant processes.
- 4. Optional paradigmatic processes and/or dyslalia
- 5. Normal phonology and articulation

The classification of stages was made by the author of this thesis (I.L.). Data were expressed with descriptive statistics for the demographics. Differences between the groups with respect to non-parametric data were analyzed with the Mann-Whitney *U*-test. Changes in phonological development before and after surgery within the study groups were analyzed using the Wilcoxon signed-rank test. *P*-values <0.05 were considered statistically significant. A correlation analysis of the developmental stage and the result of the NOT-S was performed using Spearman's rank correlation coefficient.

All statistical analyses were performed using SPSS© Windows version 15.0 (study I and IV) and version 17.0 (study II and III). *P*-values <0.05 were considered statistically significant.

# Reliability

## Study I.

A random choice of 10% of the assessments of the children operated was video-recorded to check for inter-rater agreement. The inter-rater agreement of the video recorded assessments was 100% between the two principal investigators and SLP's (I.L. and A.McA) and 88% between the SPL's and trained master students.

#### Study II.

Inter-rater agreement across listeners was calculated with Cronbach's Alfa and found to be 0,93 A random selection of 10% of the recordings were copied and mixed with in the material in order to determine intra-rater agreement, also calculated with Cronbach's Alfa and found to be 0,99, 0,99 and 0,93 for the three listeners respectively.

## Study III.

Inter-rater agreement was calculated with Cronbach's Alfa and found to be 0.81 A random selection of 10% of the recordings were copied and mixed with the material in order to determine intra-rater agreement, also calculated with Cronbach's Alfa. The intra-rater reliability found to be 0.97, 0.73 and 0.65 for the three listeners respectively.

### Study IV.

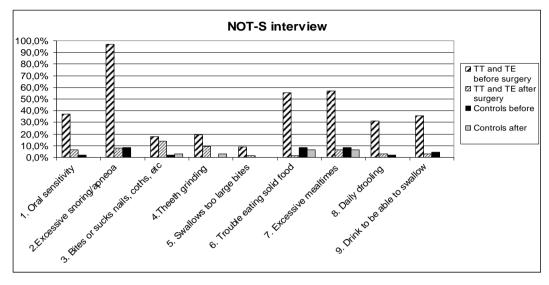
A random selection of 10 recordings were transcribed and analyzed by the third author of the study; C. S. C.S. was blinded with respect to surgical method and pre- or postoperative status. The percentage agreement between the transcribers was 96,4 %. Disagreements concerned allophonic variations. Regarding the analysis in terms of phonological processes the agreement was 100%.

## **Results**

## Study I.

## Preoperative results

At the preoperative assessment there were no differences between the TE and the TT groups in the structured interview part of the NOT-S, apart from drooling, with fewer children drooling in the TE group (p<0,5). Compared to age-matched controls, both study groups differed in all domains of the structured interview: snoring and/or sleep apnoea ("Breathing"), gag reflex elicited when brushing their teeth ("Sensory function"), teeth-grinding ("Habits"), trouble eating solid foods and excessive time spent eating ("Chewing and swallowing"), having to drink in order to be able to swallow ("Dryness of the mouth"), and drooling (p≤0.001). The item "Coughing during meals" and "Biting/sucking on fingers or objects" also differed compared to age-matched controls (p<0.01). The item "Swallowing too large bites" was more common in both study groups as compared to the age-matched controls (p<0.05), see figure 2. "Teeth-grinding" and "Drooling" were more common among boys than girls in both study groups (p<0.05), otherwise no gender differences were observed.

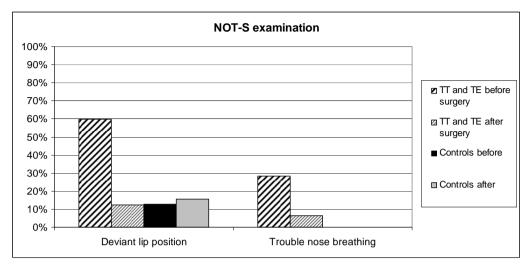


**Figure 2.** NOT-S, interview results before and after surgery for children operated and corresponding controls. Only items where differences to controls existed before surgery and postoperative changes were noted are included.

In the clinical examination part of the NOT-S there was no difference between the TE and TT groups before surgery Both the study groups differed from the age-matched controls on two items; "Deviant lip position" (Face at rest) and "Cannot keep the mouth closed and take five deep breaths" (Nose breathing) (p<0.0001), which means that they had an open mouth position. Problems with pouting and rounding the lips differed from the controls as well as use of the tip of the tongue ("Oral motor function" at p<0.01), see figure 3. No differences were seen regarding other items in the clinical examination.

#### Postoperative results

The outcome of surgery as assessed with both parts of the NOT-S was the same for both tonsillectomy and tonsillotomy. Postoperative improvements were recorded in the domains "Sensory function", "Breathing", "Chewing and swallowing", "Drooling" and "Dryness of the mouth" (p<0,0001). The study groups did not differ from the controls in any domain of the structured interview. No gender differences were observed, neither in the study groups nor in the age-matched control group.



**Figure 3.** NOT-S, examination results before and after surgery for children operated and corresponding controls. Only items where differences to controls existed before surgery and postoperative changes were noted are included.

# Study II.

## Preoperative results

The perceptual evaluations made from the preoperative recordings demonstrated that the children in the study groups had higher mean ratings on VAS for the parameters "hyponasality" (p<0,05) and "compressed/throaty" (p<0,05) and lower for "pitch" (p<0,01), see table 4 and 5. Despite randomization, scattered differences were also found between the two surgical groups. They were that the children randomized to tonsillotomy (TT) were rated to have more breathy voices (p<0,05) and the children randomized to tonsillectomy (TE) were rated to have a higher pitch. Compared to the age matched controls, the TE-group did not differ on any parameter but the TT-group had higher ratings on VAS for the parameters "hyponasality" and "compressed/throaty voice" (p<0,05). The controls had higher ratings of pitch (p<0,01) than the TT-group, see table 4 and 5.

**Table 4.**Perceptual analysis of the parameters hoarseness, breathiness, hyperfunction, hyponasality, hypernasality, roughness and compresses/throaty using VAS for study groups and agematched controls preoperatively expressed in average scores.

materied controls preop	TE+TT	Control	P- value*	TE vs TT	P- value*
Hoarseness a)	20.83±1.9	18.45±2.8	ns	19.33±2.7/22.2±2.9	ns
Breathiness a)	24.36±2.2	22.59±3.1	ns	19.75±2.8/28.82±3.2	0.035
Hyperfunction <sup>a)</sup>	14.86±1.2	17.9±2.3	ns	16.6±1.8/13.13±1.2	ns
Roughness	6.5±0.8	5.7±0.9	ns	6.9±1.3/6.15±0.9	ns
Hyponasality <sup>a)</sup>	5.06±0.7	2.43±0.6	0.011	3.91±0.6/6.17±1.6	ns
Hypernasality <sup>a)</sup>	1.19±0.2	1.24±0.3	ns	1.13±0.2/1.26±0.2	ns
Compressed/throaty <sup>a)</sup>	10.51±1.2	7.31±1.4	0.038	7.95±1.3/12.98±1.9	ns

VAS= Visual Analogue Scale (0-100 mm), TE= Tonsillectomy TT=Tonsillotomy

**Table 5.**Perceptual analysis of pitch using VAS for study groups and age-matched controls pre- and postoperatively expressed in average scores.

	TE+TT	Control	P-	TE vs TT	P-
	n=67	n=57	value*	n=33/n=34	value*
Pitch preoperatively	99.2±1.0	103.6±1.4	0.009	101.74±1/96.74±1.5	0.027
Pitch postoperatively	100.88±0.8	104.48±1.6	ns	100.57±1.1/101.18±1.3	ns

VAS= Visual Analogue Scale (0-200 mm), TE= Tonsillectomy TT=Tonsillotomy

The acoustic analyses did not demonstrate any significant differences between the TE- and TT-group at the preoperative assessment. Compared to the age matched controls, the children in the study-groups together had higher values on jitter (p<0,05), shimmer (p<0,01) and Noise to Harmonics Ratio (NHR) (p<0,001) for all vowels, se table 6.

<sup>&</sup>lt;sup>a)</sup>Mean±SD, \*Mann Whitney U-test

a)Mean±SD, \*Mann Whitney U-test

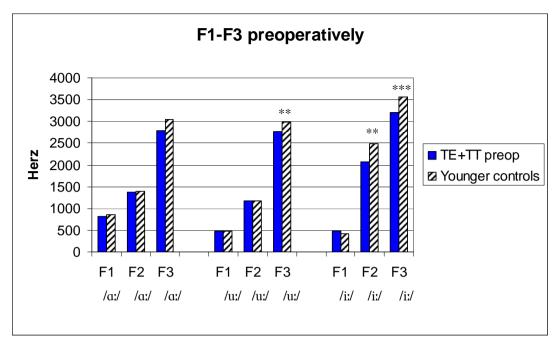
**Table 6.** Mean vocal parameters values: Jitter percent, shimmer and noise to harmonic ratio (NHR) for study groups and controls preoperatively

vowel

	/a/				/u/		/ <b>i</b> /			
Parameter	TE+TT	controls	P- value*	TE+TT	controls	P- value*	TE+TT	controls	P- value*	
Jitter a)	1.11 ±0.18	0.57 ±0.05	<0.05	1.14 ±0.17	0.65 ±0.06	<0.05	1.39 ±0.18	0.72 ±0.1	<0.05	
Shimmer a)	13.52 ±1.12	7.95 ±0.98	<0.01	14.10 ±1.19	6.39 ±0.92	< 0.001	13.88 ±1.08	6.04 ±0.72	<0.001	
NHR a)	0.14 ±0.03	0.05 ±0.01	<0.001	0.11 ±0.15	0.02 (±0.01)	<0.001	0.13 ±0.02	0.03 ±0.01	<0.001	

TE= Tonsillectomy, TT=Tonsillotomy, <sup>a)</sup>Mean ±SD, \*Mann Whitney U-test

The study groups did not differ from the controls regarding fundamental frequency (f0). Regarding formant frequencies the study groups had lower F3 values for /u/ and /i/ (p<0,01 and 0,001). For /i/, a lower F2 value was also found (p<0,001), see figure 4.



**Figure 4.** Center frequencies for formant 1-3 in the two study-groups before surgery and corresponding controls (TE=tonsillectomy, TT=tonsillotomy).

## Postoperative results

There were no significant differences between the perceptual ratings of the voices of the two operational groups and there were no significant differences between the children in the study groups and the corresponding controls. When comparing the ratings of the two study groups before and after surgery, differences were found in both study groups. The perceptual ratings of the voices in the TE-group were lower for *roughness* (p<0,05) and higher for *breathiness* (p<0,01) after surgery. For the voices of the children in the TT-group *roughness* was also lower (p<0,01) as well as *compressed/throaty* (p<0.001), but higher concerning *pitch* (p<0,01), see table 7.

**Table 7.** Perceptual analysis of the parameters hoarseness, breathiness, hyperfunction, hyponasality, hypernasality, roughness, compresses/throaty and pitch using VAS for study

groups pre- and postoperatively expressed in average values.

	TE preop/	Р-	TTpreop/	P-	TE+TTpre/	Р-	
	TE postop	value*	TT postop	value*	TE+TT post	value*	
	n=33	value	n=34	value	1E+11 post	value	
11	19.33±2.7/		22.28±2.9/		20.83±1.9/		
Hoarseness	22±2.8	ns	22.28±2.9	ns	$22.14\pm2$	ns	
Breathiness	19.75±2.8)/	0.008	28.82±3.2/	ns	24.36±2.2/	ne	
Breammess	28.14 (3.3)	0.008	28.61±3.3	115	$28.38\pm2.3$	ns	
Han out a sti ou	16.6±1.8)/		13.3±1.2/		14.86±1.2/		
Hyperfunction	$15.62\pm2$	ns	12.57±1.6	ns	$14.07 \pm 1.2$	ns	
Danahaasa	6.9±1.3)/	0.016	6.15±0.9)/	0.002	$6.5\pm0.8$	0.0001	
Roughness	$4.56 \pm 0.8$	0.010	4.21±0.9	0.002	$4.38 \pm 0.6$	0.0001	
Ummanagalitu	3.91±0.6/	***	6.17±1.6)/	0.001	$5.06\pm0.7$	0.0001	
Hyponasality	2.43±0.6	ns	2.31±0.6	0.001	$2.37\pm0.6$	0.0001	
Hypernasality	1.13±0.2/	ne	$1.26\pm0.2$	ne	$1.19\pm0.2$	ne	
пуретназанну	$1.67\pm0.4$	ns	$1.2\pm0.2$	ns	$1.43\pm0.2$	ns	
Compressed	7.95±1.3/		12.98±1.9/	0.002	10.51±1.2/	0.002	
/throaty	6.64±1.3	ns	$7.45 \pm 1.4$	0.002	$7.05\pm0.9$	0.003	
Pitch	101.74±1.5/	20	96.74±1.5/	0.003	99.2±1/	0.047	
1 исп	$100.57 \pm 1.1$	ns	101.18±1.3	0.003	$100.88 \pm 0.8$	0.047	

VAS= Visual Analogue Scale (0-100 mm. 1-200 for pitch), TE= Tonsillectomy TT=Tonsillotomy,

The acoustic analyses showed a decrease in almost all measures of perturbation for the study groups after surgery with a slight difference between the two study groups. The children in the TT-group had higher shimmer value on the vowel /u/ (p<0,05) and higher NHR for /a/ and /u/ (p<0,05) compared to the children in the TE group, see table 8.

a)Mean±SD, \*Wilcoxon signed rank test

**Table 8.** Mean vocal parameters values: Jitter percent, shimmer and noise to harmonic ratio (NIJP) for study groups postopostively.

(NHR) for study groups postoperatively.

Vowels

	/a/				/u/		/i/		
Parameter	TE	TT	P-value*	TE	TT	P-value*	TE	TT	P- value*
Jitter <sup>a)</sup>	0.86 ±0.14	0.82 ±0.11	ns	1.0 ±0.13	1.25 ±0.20	ns	1.12 ±0.24	1.53 ±0.30	ns
Shimmer a)	1.29 ±1.05	14.16 ±1.1	ns	10.37 ±1.16	15.40 ±1.49	<0.05	10.52 ±1.32	14.34 ±1.44	ns
NHR a)	0.11 ±0.02	0.15 ±0.02	< 0.05	0.05 ±0.01	0.13 ±0.02	ns	0.08 ±0.02	0.13 ±0.02	ns

TE= Tonsillectomy. TT=Tonsillotomy, a)Mean ±SD, \*Mann Whitney U-test

When the two study groups together were compared to the older controls higher values were found for all perturbation measures for the study groups (p<0,01), see table 9.

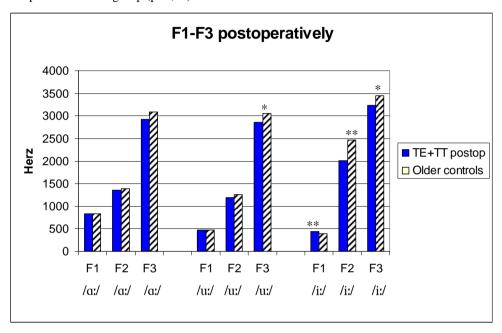
**Table 9**. Mean vocal parameters values: Jitter. shimmer and noise to harmonic ratio (NHR) for study groups postoperatively compared to age-matched controls.

Vowels

	/a/				/u/		/i/		
Parameter	TE+TT	Controls	P-value*	TE+TT	Controls	P-value*	TE+TT	Controls	P- value*
Jitter a)	0.84	0.55	< 0.01	1.14	0.51	< 0.001	1.33	0.53	<0.001
Jitter "	±0.06	±0.03	<0.01	±0.08	±0.03	<0.001	±1.2	±0.03	<b>\0.001</b>
Shimmer a)	12.62	6.3	<0.001	13.02	4.8	< 0.001	12.47	4.9	<0.001
Shimmer "	±5.1	±2.9	< 0.001	±6.4	±2.9	<0.001	±6.5	±2.4	<0.001
NHR <sup>a)</sup>	0.13	0.04	c0.001	0.09	0.01	<0.001	0.10	0.02	<0.001
NHR "	±0.1	±0.03	<0.001	±0.09	±0.01	<0.001	±0.1	±0.02	<0.001

TE= Tonsillectomy. TT=Tonsillotomy, <sup>a)</sup>Mean ±SD \*Mann Whitney U-test

The analyses of center frequency of formant 1-3 showed significant differences in comparison to age-matched the controls for the /i/-sound (p<0,01, 0,01, 0,05 respectively). The difference seen regarding the lower third formant (F3) of the /u/-sound still remained, see figure 5. The only difference between the two surgical groups was a higher F3 of /i/ in the TE group compared to the TT group (p<0,05).



**Figure 5**. Fundamental frequency and center frequencies for formant 1-3 in the two studygroups after surgery and age-matched controls (TE=tonsillectomy, TT=tonsillotomy).

There were no significant differences between the younger and the older controls.

#### Gender differences

An overview of gender differences is shown in table 10. In the study groups preoperatively, perceptual evaluation of the boys voices showed higher values for *hoarseness* (p<0,05) and the girls were rated to have higher *pitch* (p<0,05). In the younger control group the girls were rated to have morse *hoarse* voices than the boys (p<0,05). Postoperatively the only gender difference was that the girls in the study group were rated to have more *high pitched* voices than boys (p<0,05).

**Table 10**. Significant gender differences in study group pre and postoperatively and in the younger and older controls.

	♀/♂ TE+TT preop	P-value*	♀/♂ Younger controls	P- value	♀/♂ TE+TT postop	P-value*
Hoarseness a)	15,69±2,4/ 24,71±2,8 101,93±1,7/	0,05	23,68±4,8/ 14,84±3,4	0,05	 106,03±1,3/	ns
Pitch a)	97,33±1,3	0,05		ns	97,36±1,0	0,001
Shimmer <sup>a)</sup> /u/		ns		ns	15,73±1,6/ 10,83±1,2	0,05
NHR <sup>a)</sup> /a/		ns		ns	0,16±0,02/ 0.11±0,02	0,05
NHR <sup>a)</sup> /u/		ns		ns	0,13±0,03/ 0,06±0,01	0,05
NHR <sup>a)</sup> /i/		ns		ns	0,14±0,03/ 0,07±0,02	0,05
Formant 1 a) /u/	520±17,2/ 446±18,3	0,05		ns		Ns
Formant 2 a) /a/		ns		ns	1477±50,7/ 1288±31,6	0,01

 $TE{=}\ Tonsillectomy,\ TT{=}Tonsillotomy,\ ^a{)}Mean\ Mean{\pm}SD,\ *Mann\ Whitney\ U{-}test$ 

No gender differences were seen regarding perturbation measures preoperatively, but a single difference in formant 1 (F1) with higher mean frequency for girls in the study group. Postoperatively, girls in the study group had higher shimmer values for the /u/- sound

(p<0,05), higher NHR for all three vowels (p<0,05) and also a higher F2 value for /a/. No gender differences regarding acoustics were seen in the controls.

#### Study III.

#### Preoperative results

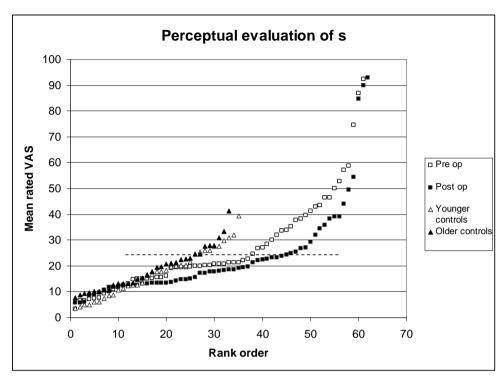
The perceptual analysis of the /s/-sound at the preoperative assessment did not show any difference between the tonsillectomy- and the tonsillotomy-group. Compared to the age-matched controls, the tonsillectomy- and tonsillotomy-groups together differed (p<0.01) with higher mean ratings of deviations on VAS, see table 11. No gender differences were seen in the study groups or in the younger controls.

**Table 11.**Perceptual analysis of /s/-articulation with VAS for study groups and controls expressed in mean values and standard deviations.

	Tonsillectomy+tonsillotomy	Controls	P-
			value*
Preop Tonsillectomy+Tonsillotomy/younger	27.2±18.8 <sup>a)</sup> n=61	17.3±9.3 a) n=35	<0.01
control group  Postop	11-01	n=33	
Tonsillectomy+Tonsillotomy/older	23.5±18.2 a)	19.8±10.6 a) n=34	ns
control group	n=62	11-34	

VAS= Visual Analogue Scale (0-100 mm) <sup>a)</sup>Mean±SD \*Mann Whitney U-test

Following a procedure developed by Sederholm et al. (Sederholm, McAllister, Sundberg, & Dahlqvist, 1993), the mean ratings for each child was plotted in rank order for each group separately. The graphs exhibit a discontinuity (an 'elbow') at approximately 25 mm VAS for all groups, see figure 6. This discontinuity served as an operational definition with deviant /s/sound production above the borderline. According to this definition 39% of the study group before surgery and 27% after surgery had deviant /s/-sounds, compared to 25.7% of the younger controls and 17.6% of the older controls, see figure 7. When comparing this borderline with the SLP's overall deviancy-ratings, all children above 25 mm VAS but four were rated as deviant.



**Figure 6.** Mean VAS-ratings plotted in rank order. The dotted line indicates the discontinuity at approximately 25 mm.

The acoustic analysis did not demonstrate any significant differences between the tonsillectomy- and the tonsillotomy-groups at the preoperative assessment. Compared to the age-matched controls, the children in the study-groups together differed significantly (p<0.05) with longer noise duration in word final position and lower peak location-values in word initial and medial position, see table 12 and 13.

#### Postoperative results

The statistical analysis of the perceptual evaluation six months after surgery did not show any significant improvement compared to the preoperative ratings. The children operated did not differ from the age-matched controls, neither as a whole group nor when divided according to surgical methods. There were no significant differences between the assessments of the /s/sounds of the children operated with tonsillectomy and the children operated with tonsillotomy, see table 11.

There were no significant differences between the younger and the older control groups.

Regarding the acoustic analyses the study groups differed from the older control group at the postoperative assessment with longer noise duration in all positions and lower peak location value in word medial position. The same difference was seen with the younger controls having longer noise duration and higher peak locations than the older control group, see table 12 and 13. A longer noise duration was seen in girls compared to boys in the study groups (p<0.05). The results were not affected by excluding the children with absent incisors.

There was no significant difference between the two study groups regarding noise duration or peak location. There were no significant differences between the preoperative and postoperative values for the study groups.

**Table 12.**Noise Duration values for /s/ in word initial, medial and final position for study groups and controls.

controls.	ND initial sec <sup>a)</sup>	P- value	ND medial sec <sup>a)</sup>	P- value	ND final sec a)	P- value
TE/TT preop	0.17±0.02/ 0.13±0.01	ns*	0.23±0.02/ 0.22±0.02	ns*	0.17±0.02/ 0.20±0.02	ns*
TE+TTpreop/ younger controls	0.15±0.01/ 0.13±0.01	ns*	0.22±0.01/ 0.24±0.01	ns*	0.20±0.02/ 0.15±0.01	<0.01*
TE/TT postop	0.13±0.01/ 0.12±0.01	ns*	0.21±0.01/ 0.2±0.01	ns*	0.17±0.01/ 0.17±0.01	ns*
TE+TT preop/ TE+TT postop	0.15±0.01/ 0.12±0.01	$ns^\nabla$	0.22±0.01/ 0.20±0.01	$ns^\nabla$	0.20±0.02/ 0.17±0.01	$ns^\nabla$
TE+TT postop/ older controls	0.12±0.01/ 0.10±0.01	<0.05*	0.20±0.01/ 0.16±0.01	<0.01*	0.17±0.01/ 0.11±0.01	<0.0001*
Younger controls/ older controls	0.13±0.01/ 0.10±0.01	<0.01*	0.24±0.01/ 0.16±0.01	<0.0001*	0.15±0.01/ 0.11±0.01	<0.05*

TE= Tonsillectomy TT=Tonsillotomy <sup>a)</sup>Mean±SD \*Mann Whitney U-test <sup>\(\nabla\)</sup>Wilcoxon's signed rank test

**Table 13.**Peak Location- values (Hz) for /s/ in initial, medial and final position for study groups and controls expressed in average scores

	PL initial	P-	PL medial	P-	PL final	P-
	Hz a)	value	Hz a)	value	Hz a)	value
TEC /TET	6154±618/	ns*	6674±598/	ns*	5635±583/	ns*
TE/TT preop	6674 ±667	118	6087±529		6297±787	
TE+TT preop /	6450 <u>±</u> 446/	<0.05*	6374±396/	-0.01*	6027±519/	ns*
younger controls	7673±356	<0.05*	7761±341	<0.01*	6658±359	
TE / TT postop	6763±404/	*	6912 ±546/	v	5293±428/	ns*
	5923±629	ns*	7477±494	ns*	5739±391	
TE+TT preop /	6450±440/	$ns^ abla$	6374±396/	$ns^\nabla$	6027±519/	$ns^\nabla$
TE+TT postop	6343±375	ns	7229±365	ns	5527±287	ПS
TE+TT postop /	6343±375/	*	7229±665/	-0.0001*	5527±287/	ns*
older controls	5134±397	ns*	5255±352	<0.0001*	4946±308	
Younger controls/	7673±356/	۰0.001*	7761±341/	-0.0001*	6658±359/	۰0.01 <b>*</b>
older controls	5134±397	<0.001*	5255±352	<0.0001*	4946±308	<0.01*

TE= Tonsillectomy TT=Tonsillotomy a Mean±SD \*Mann Whitney U-test VWilcoxon's signed rank test

A significant negative correlation (Spearman's rho -0,405) was found between the perceptual ratings and the peak location values of the study groups (p<0.001). No significant correlation was found between perceptual ratings and noise duration.

## Study IV.

#### Preoperative results

There were no differences between the TE and TT groups in terms of phonological development before surgery (ns). A majority of these children (42/67, 62.7 %) showed a slowed phonological development (developmental stages 0-4), compared to the age-matched control group, where 34% remained on stages 0-4 (p<0.001), see table 14 and figure 7. Two children in the tonsil group (3 %) were at developmental stage 2 with pervasive reduction of more than one type of cluster, optional assimilation, and/or deletion of pretonic syllables. None of the controls were at stage 2. Seventeen children in the tonsil group (25.4 %) and two (4.3%) of the age-matched controls were at developmental stage 3, i.e. predominantly reduction of s-clusters and pervasive paradigmatic processes. Twenty-three children in the tonsil group, (34.3 %) and 14 age-matched controls (29.7%) were at developmental stage 4, i.e. optional paradigmatic processes and/or dyslalia. Twenty-five children (37.3%) in the

tonsil group and 32 (68.1 %) of the controls were assessed to have fully developed phonology. The difference in degree of developmental stages between the tonsil groups and age-matched controls was statistically significant (p<0.001).

 Table 14. Categorization of the children's phonological development into developmental

stages, expressed in percentage (number of).

Developmental stage	TE before surgery	TT before surgery	Corresp.	TE after surgery	TT after surgery	Corresp.
	n=33	n =34	n =47	n =32	n =32	n =31
2	6.1 (2)	0	0	6.2 (2)	0	0
3	27.3 (9)	23.5 (8)	4.2 (2)	18.8 (6)	12,5 (4)	0
4	30.3 (10)	38.2 (13)	27.7 (13)	34.4 11)	25 (8)	3.2 (1)
5	36.4 (12)	38.2 (13)	68.1 (32)	40,6 (13)	62.5 (20)	96.8 (30)

TE= Tonsillectomy TT=Tonsillotomy

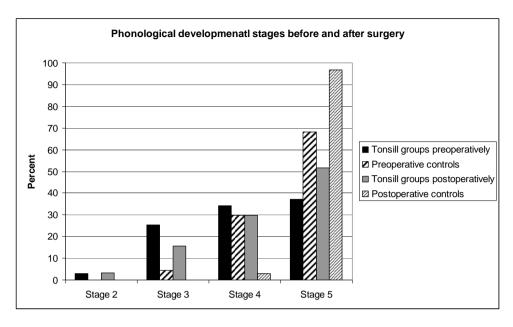
#### Postoperative results

The children in both the tonsil groups had improved their phonological skills by the time of the postoperative assessment (p<0.01). However, the difference between the children in the tonsil group and the postoperative control group was more pronounced than before surgery. Among the children in the tonsil group 31/65 (48.4%) had phonology at developmental stages 2-4 compared to just one child out of 32 (3.1 %) in the age-matched control group, (p<0.001) at stage four, see figure 7.

After surgery there were further phonological improvements among children who received TT than TE (p<0.05), see table 13.

There was no relationship between the results from the phonological analyses and those from the oral-motor assessments using NOT-S neither before nor after surgery. Similarly, there was no relationship between these assessments in the controls.

There was no relationship between the phonological development in the children with otosalpingitis (OME) and grommet insertion.



**Figure 7.** Results from the phonological assessment before and after surgery for children operated and corresponding controls.

### **Discussion**

The results of the thesis project clearly show that tonsillar hypertrophy influence several different aspects of the speech and language spectrum. These aspects in turn have great impact on general well-being and quality of life (Richards & Giannoni, 2002; Fujiki, Spackman, Brinton & Hall, 2004; Damiano, Tyler, Romitti, Momany, Jones, Canady, Karnell, & Murray, 2007). Children with tonsillar hypertrophy and their caregivers have reported a reduced quality of life (Ericsson, Lundeborg et al., 2009). Despite these findings, problems of voice, speech and language in connection with tonsillar hypertrophy have earlier not been recognized as additional indications for surgery. However, there is some evidence that oral motor problems may influence the treatment decision process (van den Akker et al., 2003). Almost all children studied had oral-motor problems in addition to the breathing obstruction. The problems did not only impact the child but often the whole family. For many parents, mealtimes had been a struggle with a child not wanting to eat and in particular, not wanting to swallow solid food. The parents frequently volunteered information that the whole family had grown tired of waiting for the child to finish their meal. A shared mealtime offers great opportunity for interaction and communication and by participating the child learns social

skills connected with family and communication (Evans Morris & Dunn Klein, 2000). Another oral-motor problem found preoperatively in the study group was drooling. In a child's normal development, the control of drooling gradually increases during the first two years of life (Evans Morris & Dunn Klein, 2000). If drooling persists in children aged four years and older, psycho-social and physical well-being can be negatively affected (Nunn, 2000).

Voice function was clearly affected in the study group before surgery with significantly higher ratings of the parameters, *hyponasality* and *compressed/throaty voice* in comparison to age-matched controls. Several studies have shown that listeners rate children with voice disorders as having more negative personal traits compared to controls (Ruscello, Lass & Podbesek, 1988, Lass, Ruscello, Bradshaw, & Blankenship, 1991; Lass, Ruscello, Stout & Hoffman, 1991).

The /s/-sound is one of the most common consonant phonemes in Swedish. When a prevalent phoneme is distorted, intelligibility may be compromised. In this project children with tonsillar hypertrophy were assessed as having significantly poorer /s/-articulation than age-matched controls. Many children in the study group preoperatively, also had a deviant lip position with an open mouth indicating oral breathing. At the postoperative assessment some of them (12%) still had this open mouth position. This may be explained as a well established habit. It is of importance for these children to change this open mouth habit in order to improve the defective /s/-articulation and avoid a negative orthodontic and facial development (Guilleminault, Partinen, Praud, Quera-Salva, Powell & Riley, 1989; Zettergren-Wijk, Forsberg, & Linder-Aronsson, 2006). Also, teachers' as well as peers' attitudes towards children with deviant articulation have been shown to be more negative (Ruscello, Stutler, & Toth, 1983; Crowe Hall, 1991; Overby, Carell & Bernthal, 2007).

A new and surprising finding of this study was the phonological problems in the study group. The disturbed sleep in OSDB and the subsequent effects on cognitive functioning may be an explanation. Perception, cognition, verbal short-term memory, sensory function, and motor control of structures in the mouth and vocal tract are considered to be relevant factors for language development (Grunwell, 1997). All these factors are affected by tonsillar hypertrophy (Ahlqvist-Rastad et al., 1988; Messner & Pelayo, 2000; Chervin et al., 2005). Despite the common knowledge of the impact of these factors the difference in phonological development between the study group and age-matched controls was more pronounced than expected. Since this a result previously not reported, the rational for including a phonological assessment was primarily correlation with oral motor dysfunction. However this correlation

was not found. An alternative explanation to the findings regarding poor phonological development could be that the hampered oral motor function influenced the phonological development during a critical period. This shows that the simple explanation is not always valid. Co-occurrence of more than one problem is not uncommon. The children in the study group are clear examples of this with speech, language, voice and oral motor problems preoperatively. The interdependence between these functions needs to be further elucidated from a developmental perspective.

Phonology appeared to be improved at the postoperative assessment but the development was not fast enough to catch up. Actually, the gap between the children and the age matched controls was larger at postoperative assessment. This finding supports the "critical period hypothesis" with a developmental window for optimal language acquisition, said to be "open" until approximately four years of age (Ruben, 1997). It is therefore imperative to provide speech and language therapy for children with tonsillar hypertrophy as early as possible and not wait until after surgery.

The problems studied in relation with tonsillar hypertrophy and OSDB were equally improved after TE and TT. Since several studies have shown that tonsillotomy (TT) gives less surgical trauma (less postoperative pain and shorter recovery time) this procedure can be recommended.

Some methodological issues must be addressed.

Thirty-seven families out of 118 invited, declined enrolment in the study, which is to be expected when the randomization is performed prior to the invitation to the study (Zelen, 1981). Since the number of children who did not was about the same for TE and TT, no extra analysis of them as a group was regarded necessary. However, the proportion of children with problems with functions studied might have been slightly higher among the participants than among the children who were not enrolled, reflecting a greater interest in the study for families experiencing the problems to be investigated (Murphy, Escamilla, Blackwell, Lucke, Miner-Williams, Shaw & Lewis, 2007).

For all four studies, different children have been used as age-matched controls pre- and postoperatively. Ideally the same children should have been assessed two times and for all studies. However, the assumption is that the groups of "typically developed" children are representative for the given assessment regardless of age.

The children of the study group were exposed to several assessments during the same visit. Besides the assessment made for this study, an ENT-examination, an orthodontic examination including taking of dental casts were performed at the preoperative visit. In order to minimize discomfort for the children the number of tests was limited. For this reason formal hearing tests and sleep assessments were not included. Instead a thorough patient history including questions about hearing was made during the preoperative ENT examination. Only a small number of children (six, equally distributed between the two surgical groups) were in need of grommet insertion. These children did not differ from the other children in the study group. Thus there were no bases to address the question about to what extent OME influences speech development.

Apart from those made for the inter-reliability testing, no video-recordings were made of the assessments. Perhaps the outcome of the oral motor examination would have been different if such recording had been made.

In this study relatively young children participated. Before surgery they had never met the person collecting the speech material and the recordings were made in a non-familiar setting. Some of them refused to complete all the material. Others participated, but whispered or shouted which made the acoustic analyses impossible.

## **Conclusions and clinical implications**

The overall aim of this thesis was to evaluate four different functional aspects of communication in preschool children with tonsillar hypertrophy and to study the effects of two types of tonsil surgery, tonsillectomy or tonsillotomy combined with adenoidectomy when necessary.

Compared to age matched controls, children with tonsillar hypertrophy have:

- oral motor dysfunction as measured by the Nordic Orofacial Test-Screening(NOT-S),
- perceptually and acoustically deviant voices compared to controls
- perceptually and acoustically deviant /s/-articulation and,
- a higher prevalence of and more severe phonological deficits

Six months after surgery the oral-motor problems and voice quality were normalized. Other studied functions (s-articulation and phonology) were better, but not enough to match controls. Regarding the phonological development, the gap compared to controls increased.

Both SLP's and ENT-surgeons need to be aware of the potential impact of tonsillar hypertrophy on oral-motor function and the speech and language spectrum to be able to help

affected children adequately. SLP's must consider that speech and language problems can be caused by different underlying factors where tonsillar hypertrophy is one. A simple screening of oral structures and motor functions should be included in the pediatric SLP's routine. Communication and cooperation between SLP's and surgeons is crucial for adequate assessment and care of children with obstructive breathing. Surgeons should refer affected children to SLP's in order to be able to include oral motor and speech and language problems in the decision process for tonsillar surgery. And also the reverse, SLP's should refer children with anamnestic or clinical findings of obstructed breathing to ENT-surgeons.

The outcome of the two surgical methods was the same for all studied functions. Since tonsillotomy is associated with less postoperative morbidity and pain it should be the preferred method.

# **Svensk sammanfattning (Swedish summary)**

I föreliggande avhandlingsarbete studeras förekomst orofacial funktion, röstfunktion, artikulation och språkfunktion hos barn med tonsillförstoring samt hur dessa funktioner påverkas av två olika typer av kirurgisk behandling.

De flesta barn i förskoleåldern har en relativ förstoring av lymfatisk vävnad i nässvalget som ett direkt immunologiskt svar på alla de infektioner de exponeras för. Hos många av dessa barn förekommer obstruktiva besvär och andra associerade symptom av varierande grad. De vanligaste rapporterade symptomen är snarkning, orolig sömn och andningsuppehåll nattetid och trötthet, koncentrationssvårigheter, munandning, dregling, ätsvårigheter, grötig tal och påverkad röstkvalitet dagtid. Den behandling som erbjuds barn med obstruktiva besvär är kirurgi i syfte att minska storleken av tonsillvävnaden. I Sverige utförs årligen cirka 10 000 tonsilloperationer. Två olika kirurgiska metoder används vid tonsilloperation. Den vanligast förekommande metoden är tonsillektomi, TE, varvid hela tonsillen och tonsillkapseln avlägsnas. Metoden är förknippad med en hel del smärta efter operationen och det föreligger också en risk för blödningskomplikationer. En alternativ metod, tonsillotomi (TT), vid vilken endast den obstruktiva, utskjutande delen av tonsillvävnaden avlägsnas, har under den senaste tioårsperioden åter introducerats i Sverige. Ett flertal studier har visat att metoden är skonsammare för barnet med lägre blödningsrisk och väsentligt lägre smärtpåverkan i efterförloppet samt snabbare återgång till barnomsorg samtidigt som metoden har samma positiva långtidseffekter beträffande sömnstörning och infektionsbenägenhet. Frågan är om

tonsillotomi ger samma positiva långtidseffekter för andra funktionella besvär som är kopplade till tonsillhypertrofi.

Sextiosju barn i åldrarna 4;5 -5;5 år, undersöktes i föreliggande avhandlingsarbete. De rekryterades ifrån väntelistorna för tonsillkirurgi vid tre Öron-Näsa-Halskliniker i Sydöstra Sjukvårdsregionen och slumpades till antingen traditionell tonsillektomi eller tonsillotomi. Inför operation undersöktes och ljudinspelades samtliga barn med avseende på orofacial motorik, röst, artikulation och språkljudsorganisation (fonologi). Samma undersökning genomfördes igen sex månader efter operation. Tvåhundrasex friska förskolebarn med typisk utveckling i samma åldrar som studiebarnen undersöktes och utgjorde jämförelsematerial till studiebarnen.

Avhandlingen består av fyra delarbeten.

I delarbete I undersöktes orofacial funktion med hjälp av ett orofacialt test, NOT-S (Nordiskt Orofacialt Test-Screening). Resultaten visade att barnen med tonsillhypertrofi hade andningspåverkan, ät- och sväljproblem, dregling och avvikande munposition (öppen mun) i väsentligt högre grad än de friska kontrollbarnen. Problemen normaliserades efter operation och effekten var oberoende av operationsmetod.

I delarbete II bedömdes röstfunktion med såväl perceptuell (lyssnarpanel) som akustisk analys. Rösterna hos barnen med tonsillhypertrofi uppfattades i högre grad vara präglade av hyponasalitet (sluten nasalitet) samt uppfattades som mera klämda eller halsiga. Den akustiska analysen visade att deras röster innehöll fler tecken på oregelbundna stämbandsvibrationer och ofullständig stämbandsslutning än kontrollgruppbarnens röster. Röstfunktionen förbättrades av operation ungefär likvärdigt för de båda metoderna, men en viss skillnad relativt de friska barnen kvarstod.

I delarbete III undersöktes artikulationen av /s/-ljudet som bedömdes perceptuellt och analyserades akustiskt. Artikulationen av /s/ hos barnen med tonsillhypertrofi bedömdes som sämre hos barnen med tonsillhypertrofi före operation i jämförelse med kontrollbarnen vilket bekräftades av den akustiska analysen. Resultaten efter operation var likvärdiga för båda operationsgrupperna och /s/-ljuden bedömdes inte skilja sig från kontrollbarnens /s/-artikulation. Akustiskt var de opererade barnens /s/-ljud mer högfrekventa än kontrollbarnens. Resultaten påverkades inte av att barn som tappat framtänder inte räknades med.

I delarbete IV undersöktes språkljudsorganisation (fonologi), som är en aspekt av språklig utveckling. Barnen med tonsillhypertrofi hade före operation en påverkan på språkljudsorganisationen så till vida att de hade såväl högre förekomst av och mer grava

fonologiska problem än kontrollbarnen. Efter operation minskade problemen med språkljudsorganisationen lika mycket för båda operationsgrupperna, men avståndet till kontrollbarnen ökade.

Sammanfattningsvis har tonsillhypertrofi stor inverkan på studerade funktioner och därmed bör indikationerna för operation vidgas till att omfatta även oralmotorik och olika aspekter av tal- och språk. Det är viktigt att barnlogopeder uppmärksammas på att tonsillhypertrofi kan finnas med som en bidragande orsak till svårigheter hos barn och att de tar initiativ till att drabbade barn får hjälp. Eftersom resultaten av de båda kirurgiska metoderna var likvärdiga, är tonsillotomi, som innebär avsevärt mindre smärta och sjuklighet hos barnen postoperativt, att föredra.

## Acknowledgements

There are several people without whose help this thesis would not have been completed and whom I want to thank.

I owe my deepest gratitude to my two supervisors, Elisabeth Hultcrantz and Anita McAllister. I started off with Elisabeth as my first supervisor and Anita as my second. After half-time, Anita became my first supervisor and Elisabeth my second. This arrangement has been most beneficial for me since both have generously shared their extensive knowledge in their different professional fields. The fact that they both are married to Americans has been an extra bonus. When in doubt about choice of words and expressions Ed and Bob have been a great help, thank you, Ed and Bob!

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# **Appendix**

# Appendix 1

Voice no:(Röst nr)	Rater:(Bedömare)	
Voice quality (Rös	kvalitet)	
Not at a		A lot/consistent (Mkt/genomgående)
Hoarseness (Intryck av heshet)		
Breathiness (Läckage)		
Hyperfunction _ (Hyperfunktion/press)		
Roughness (Skrovlighet)		
Hyponasaliy (Hyponasalitet)		
Hypernasality (Hypernasalitet) Compressed/		
Throaty (Klämd/Halsig)		
läge)	Expected (Förväntat)	H (F

Low (Lågt)

Perceptual evaluation of the s-sound.	
Rater:	
	Deviant manner
	(Avvikelse
	betr

Appendix 2

A sharp/distinct s-sound

(Skarpt/tydligt s-ljud)

No (nr)

artikulationssätt)

A very deviant/indistinct

(Mkt avv/ otydligt s-ljud)