

**Attitude to Speech and
Communication in Individuals
Born with Cleft Lip and Palate**

Attitude to Speech and Communication in Individuals Born with Cleft Lip and Palate

Christina Havstam

**Institute of Neuroscience and Physiology
at Sahlgrenska Academy
University of Gothenburg**



UNIVERSITY OF GOTHENBURG

© Christina Havstam, 2010

ISBN 978-91-628-8168-9

Printed by: Intellecta Infolog AB
Kållerød, Sweden, 2010

Abstract

The overall aim of this thesis was to explore and describe how individuals who are born with a cleft lip and palate experience their communicative situation, how they perceive their speech and whether their views correlate with the results of speech assessments made by specialised speech-language pathologists (SLPs).

Both quantitative and qualitative methods were used. Fifty-four 10-year-olds born with a cleft involving the palate completed a Swedish version of a Communicative Attitude Test (CAT-S) and the result was compared with their parents' responses to questions about their child's communicative situation and the results of speech assessments made by SLPs. The children's mean CAT-S scores were also compared with the mean score of a group of children without a cleft. Thirty-five young adults born with a unilateral or bilateral cleft lip and palate were asked about their satisfaction with speech outcome after their treatment had been terminated. They indicated their satisfaction with their speech on visual analogue scales which were compared with speech assessments. Finally, 13 young adults who had been assessed as having a speech impairment on their last visit to the cleft team participated in semi-structured interviews about their experiences of growing up with a cleft and their impressions of their speech and communication. The interviews were conducted and analysed according to the guidelines of the qualitative method Grounded Theory.

The 10-year-olds' responses to the CAT-S were significantly correlated with the majority of the speech variables, but the associations could only explain part of the variance. Their communicative attitude was significantly more negative compared with reference data from 10-year-olds without a cleft, but there were large individual differences among children born with a cleft. The adults' own satisfaction with speech was not statistically correlated with the results of the speech assessments. The analysis of the interviews resulted in two separate areas with separate core categories. The core category *Making sense of the cleft* described the processes of developing self-image in relation to the cleft and comprised the categories Shaping one's attitude to the cleft and Dealing with being different. The core category *Taking charge of communication* described the way the speech impairment was dealt with and comprised the categories Forming an idea of one's speech, Learning about one's communication and Taking responsibility for communication.

Many of the children and adults born with a cleft lip and palate who were assessed as having a speech impairment by SLPs were themselves satisfied with their speech and described well-functioning communication in most situations. Isolated speech assessments by SLPs are therefore not especially informative about the person's communicative participation in society. For this reason, it is also important to assess the persons' own attitude to their speech and communication in a structured way. Adding information about the individual's own experience would expand the field of speech-language pathology from a description of speech impairment to include communicative participation, which has implications for both clinical practice and research concerning individuals born with a cleft lip and palate.

Key words: cleft (lip and) palate, speech, communicative attitude, participation, satisfaction with speech, self-assessment, qualitative methods, grounded theory, interview, speech assessment

Contents

Abstract	5
Contents	7
List of publications	9
Background	11
Cleft lip and palate – anatomy, incidence.....	11
Speech disorders associated with clefts involving the palate.....	11
Speech in children born with a cleft palate.....	12
Speech in adults born with a cleft palate	13
Speech assessments.....	13
Treatment	15
Balancing growth and speech development	15
Speech treatment	15
Individuals’ own satisfaction with treatment outcome.....	16
International Classification of Functioning, Disability and Health (ICF).....	17
Participation in individuals born with a cleft.....	18
Communicative Participation	19
Definition.....	19
Environmental Factors – listeners’ impressions.....	20
Personal Factors.....	21
Holistic aspects of communication in quantitative research.....	21
Summary of holistic findings in quantitative research for individuals born with a cleft.....	22
Theoretical foundations in qualitative methods.....	24
Summary of findings in qualitative research for individuals born with a cleft.....	25
Aims	27
Methods	29
Participants.....	29
Children	29
Adults.....	29
Procedure in the quantitative methods.....	29
Data collection.....	29
Speech material	30
Perceptual assessments.....	30
Reliability of speech assessments	30
Statistical analyses.....	31
Procedure in the qualitative methods.....	31
Data collection.....	31
Analysis	31
Results	33
Communication attitude in children (Study I).....	33

The attitudes of children compared with the impressions of parents.....	33
Communication attitude compared with speech assessments.....	34
Satisfaction with treatment outcome in young adults (Study II).....	34
Young adults' descriptions of their cleft experience (Study III).....	35
Young adults' descriptions of their speech and communication (Study IV) ..	36
Discussion	37
Attitudes to speech and communication in children born with a cleft	37
Attitudes to speech and communication in adults born with a cleft.....	38
Growing up with a cleft	39
Dealing with a cleft-related speech impairment	40
Methods employed in the studies	41
What is the task of the cleft team?	42
What is the task of the speech-language pathologist in cleft care?	43
Limitations	44
Summary and clinical implications	44
Future research	45
Acknowledgements.....	47
References	49
Summary in Swedish.....	59

List of publications

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

- I Havstam C, Dahlgren Sandberg A, Lohmander A. Communication attitude and speech in 10-year-old children with cleft (lip and) palate in an ICF-perspective. *International Journal of Speech-Language Pathology*, 2010, in press.
- II Havstam C, Lohmander A, Dahlgren Sandberg A, Elander A. Speech and satisfaction with outcome of treatment in young adults with unilateral or bilateral complete clefts. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery* 2008;42(4):182-189.
- III Havstam C, Laakso K, Ringsberg KC. Making sense of the cleft: Young adults' accounts of growing up with a cleft and deviant speech. *Journal of Health Psychology*, 2010, in press.
- IV Havstam C, Laakso K, Lohmander A, Ringsberg KC. Taking charge of communication: Adults' descriptions of growing up with a cleft-related speech impairment. Submitted.

Reprints were made with kind permission from *International Journal of Speech-Language Pathology*, *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, and *Journal of Health Psychology*.

Background

Cleft lip and palate – anatomy, incidence

Clefts of the lip, alveolus and/or palate, orofacial clefts, have an incidence of approximately 2/1000 live births in Europe (Hagberg et al., 1997; Mossey, 2007). Orofacial clefts are the most common congenital craniofacial malformations and are due to a fusion failure in the structures forming the different parts of the mouth during the first trimester of pregnancy. Many different combinations occur and they are usually divided into 1) unilateral or bilateral clefts of the lip and/or alveolus, 2) unilateral or bilateral complete clefts of the lip, alveolus and palate and 3) isolated clefts of the palate. Furthermore, the isolated clefts are often divided into clefts involving only the soft palate (the posterior muscular part including the uvula) or both the hard (the anterior bony part) and the soft palate. Most clefts are isolated defects, but about 20% of them are part of a syndrome (Shprintzen et al., 1985).

Speech disorders associated with clefts involving the palate

The direct consequence of an open cleft in the palate is that there is an abnormal connection between the nasal and oral cavities. This means that there may be leakage of food and liquid through the cleft, as well as the passage of air and influenced resonance in speech. It is also not uncommon for the function of the Eustachian tube to be disturbed, resulting in poor ventilation of the middle ear, with an increased risk of hearing impairment. The types of speech disorder most characteristic of cleft palate speech are 1) audible nasal air escape, usually heard on high-pressure consonants (in Swedish /p/, /t/, /k/, /b/, /d/, /g/, /f/, /s/, /ʃ/, /ç/) and 2) deviant resonance, usually excessive nasal resonance, so called hypernasality, most noticeable on vowels. The opposite, insufficient nasal resonance known as hyponasality, is noticed primarily on nasal consonants, which thereby become denasalised. A combination of the two, known as mixed nasality, can also occur. Lastly, 3) weakening of the high-pressure consonants due to the inability to build up sufficient intra-oral pressure. In some cases, these difficulties persist after the cleft is surgically closed due to impaired velopharyngeal function or an unwanted opening remaining in the palate. The velopharynx is the area behind the nose where a complex muscular mechanism is activated during swallowing and production of the oral speech sounds to close off the connection between the oral and nasal cavities. This involves the upward

movement of the soft palate and a movement towards the midline of the pharyngeal walls that normally results in complete closure (Cassel & Elkadi, 1995). Due to a short or immobile soft palate, a deep pharynx or reduced motion of the pharyngeal walls, some individuals born with a cleft of the palate have velopharyngeal impairment (VPI). The incidence of VPI reported in individuals with a cleft involving the palate varies from 5% to 40% (Grunwell & Sell, 2001). VPI is also found in individuals with submucous clefts, or without any clefts, often in connection with syndromes such as the 22q11 deletion syndrome (Persson, 2004) or as a component in motor speech disorders, dysarthria (Yorkston, 1987).

The other frequent consequence of a cleft of the palate is a retracted pattern of articulation. Most consonants are normally produced by a narrowing of the vocal tract, either to complete closure as in the plosives (/p/, /t/, /k/, /b/, /d/, /g/) or to reduced passage of air as in the fricatives (/f/, /v/, /s/, /ʃ/, /ç/). When such closure or reduced passage is made impossible due to an open cleft or VPI, the affected person sometimes tries to compensate for it by placing the articulation behind the area of nasal leakage, so called compensatory, or retracted, articulation. The sound is thus produced with adequate pressure and without nasal air escape but in the wrong place in the vocal tract. The speech sound could thereby be produced as another speech sound, with a risk of making the word difficult for other people to understand. These speech disorders appear to be related to both the technique and timing of surgery (Lohmander-Agerskov, 1996). A common example of retracted oral articulation is when /t/ (in Swedish normally produced with the tip of the tongue just behind or on the back of the front teeth) is produced further back in the oral cavity; as, for example, [k]. A more severe type of disorder is retracted glottal articulation, probably caused by VPI, where the articulation is placed behind/under the velopharynx and the closure is made with the vocal cords. Nowadays, this is not a frequent disorder among people born with a cleft, but those who do develop glottal articulation are often very difficult to understand and some of them suffer from severe communication difficulties. Sometimes, the deviant patterns of articulation remain after the cleft has been closed and about 20% of the individuals born with a cleft involving the palate require long-term speech therapy due to severe problems (Grunwell & Sell, 2001).

Speech in children born with a cleft palate

When groups of babies with and without clefts in the hard palate are compared with regard to their babbling, the children with clefts produce less of the bilabial and dental/alveolar plosives heard in babies without a cleft (Jones et al., 2003; Lohmander et al., 1994; Scherer et al., 2008). During their

first years, many children born with a cleft palate continue to avoid anterior placement in the early speech production of consonants and to produce fewer high-pressure consonants (Chapman et al., 2003; Lohmander & Persson, 2008; Scherer et al., 2008). At 5-6 years of age, about 40% of the children have been reported to show signs of VPI (Lohmander & Persson, 2008; Pulkkinen et al., 2001; Sell, 2001) and some of them require secondary surgery to improve their ability to achieve sufficient velopharyngeal closure. About 5-20% of the children have also been reported to have some form of articulation disorder at 5 years of age and a survey of children born with a unilateral cleft lip and palate in the UK (Sell et al., 2001) revealed that about one-fifth of the 5-year-olds were impossible or only just possible to understand for strangers. As they grow older, speech improves for most children and the majority have normalised speech by the age of 10 years (Grunwell et al., 2000; Lohmander et al., 2006; Park et al., 2000). A minority, however, have remaining speech impairments, primarily signs of VPI but also retracted articulation at this age.

Speech in adults born with a cleft palate

Not many studies describing speech outcome in adults born with clefts of the palate exist. Their speech is often increasingly normalised during later childhood and adolescence and both children and parents have reported increasing satisfaction with speech as the children grow older (Broder et al., 1992). Some individuals, however, continue to have a speech disorder even as adults. Since studies of speech in adults are scarce, it is difficult to estimate the exact numbers. The few studies using speech recordings that have been conducted have mostly compared different surgical techniques for repairing the palate (Becker et al., 2000; Farzaneh et al., 2008; 2009; Van Lierde et al., 2004). They have reported quite different speech results, but signs of VPI constitute the most common disorder and have been reported in up to 60% of the investigated individuals. Some have found more speech deviations in bilateral than unilateral complete clefts (Farzaneh et al., 2008; 2009).

Speech assessments

Speech outcome is considered to be one of the most important criteria for evaluating the success of surgical treatment of the cleft involving the palate (Grunwell & Sell, 2001). The most common way of evaluating speech is to use listeners' subjective assessments, but there are substantive problems connected with this (Sell, 2005). Many different methods and standards have been used for collecting and analysing speech material in an international perspective and sometimes basic information, such as the reliability of listeners, is missing (Lohmander & Olsson, 2004). In studies that have reported information of this

kind, listener agreement is sometimes disappointingly low, especially for hypernasality, and this constitutes a problem when it comes to using perceptual assessments reliably (Kuehn & Moller, 2000). Efforts to find more objective methods have led to various types of instrumental assessment, such as aerodynamic measurements of pressure and flow, and acoustic measurements, such as nasometry. However, the perceptual judgement of nasality is still considered to be the most important description of speech (Kuehn & Moller, 2000; Sell & Grunwell, 2001). Moreover, the use of “blind independent analysis of speech data by specialist therapists” is recommended as the gold standard methodological approach (Sell, 2005, p. 118). One way to enhance the quality of these assessments is to calculate reliability, both comparisons between judges (inter-rater reliability) and repeated judgements by the same individual(s) (intra-rater reliability). This is commonly calculated as point-by-point (exact) agreement, or by correlation analysis or kappa statistics. Previous studies have often shown that judges agree with themselves (high intra-rater reliability) to a higher degree than with each other (low inter-rater reliability) (Lohmander-Agerskov, 1996; Persson, 2004).

Speech assessments are usually performed by rating the speech variables that are thought to describe the typical characteristics of cleft-related speech impairments. These variables are commonly hypernasality, hyponasality, audible nasal air emission or turbulence, consonant production (articulation) errors and voice disorders (Henningsson et al., 2008). It is also common to add assessments of weak-pressure consonants, which is the pressure reduction sometimes heard on the consonant usually produced with high pressure (the plosives and fricatives listed above) (e.g. Lohmander & Persson, 2008). Some studies have added a comprehensive evaluation of velopharyngeal function, comprising the variables of hypernasality, audible nasal air leakage and weak-pressure consonants (e.g. Dotevall et al., 2002; Karnell & Van Demark, 1986; Lohmander et al., 2009). The inclusion of overall variables such as speech intelligibility (understandability) and general impression of speech (or speech acceptability or naturalness) is often also recommended in order to focus on the potential consequences of a speech disorder for the affected person’s everyday life. A literature review of intelligibility measures in speakers with a cleft palate found confusion in the terminology that was used and intelligibility was not clearly separated from acceptability, severity or speech proficiency (Whitehill, 2002). Moreover, information about inter- and intra-rater reliability was missing in almost half the reviewed articles. The need for a global measure of speech performance was identified. Intelligibility is considered by many to be the most important measure of speech impairment and the most important target for intervention (e.g. Yorkston et al., 1987), but it is important to use clearly defined, reliable measures (Whitehill, 2002). It is, however, not considered to be sufficient to report intelligibility in isolation, since it is influenced by many

factors other than the actual speech characteristics due to a cleft palate, but a measure of intelligibility should be added to the report of the speech variables described above.

Treatment

Balancing growth and speech development

Historically, there has been a conflict of interest between accomplishing good speech on the one hand and good growth/appearance of the mid-face on the other (Friede, 2009). It has sometimes been a case of speech-language pathologists advocating closure of the cleft in the palate as soon as possible to achieve normalised conditions for speech development and orthodontists and surgeons wanting to postpone surgery in order not to disturb the natural growth of the face. In some cases, individuals with clefts develop mid-face retrusion and distortion of the dental arch that require secondary surgery. This is believed to be caused by both the timing and type of surgical treatment (Mars, 2001). To avoid underdevelopment of the maxilla, delayed closure of the hard palate has been in use for some 30 years at the cleft centre in Gothenburg (Lilja et al., 1996). Internationally, there are many different types of surgical treatment in use and, so far, no one method has proved to be the best treatment for repairing cleft lip and palate (Shaw et al., 2000).

Speech treatment

One of the tasks of the SLP is to help determine if the present anatomy and physiology need to be altered to make it possible to achieve normalised speech, or if the speech disorder can be amended with speech-language therapy (Sell & Grunwell, 2001). The types of treatment are briefly described below.

There are three main types of treatment for VPI.

1) Surgical alteration of the velopharynx that mainly involves two types at the cleft palate centre in Gothenburg. The first type is the velopharyngeal flap that has been used for several decades (Engström et al., 1970) and involves a flap of mucosa and muscle tissue from the posterior pharyngeal wall that is attached to the back of the soft palate, forming a bridge-like barrier between the oral and nasal cavities (Mercer & Pigott, 2001). The function presupposes sufficient lateral wall movement towards the flap and, if the flap is too broad, it may cause obstruction of the airway (usually snoring and/or hyponasal speech). The second type is the soft palate re-repair, which is a more physiologically correct operation that does not alter the normal anatomy of the pharynx. This is a procedure in which the levator muscle is moved to a more posterior position in

order to increase the functional length of the velum in order to achieve sufficient closure against the posterior pharyngeal wall (Sommerlad et al., 2002).

2) Prosthetic management, where the most common type for individuals born with a cleft palate is the speech bulb obturator. It usually consists of an acrylic plate which is fastened in the mouth with dental clasps and continues back into the pharynx where the bulb fills out the gap between the soft palate and the pharyngeal walls so that sufficient velopharyngeal closure can be obtained (Peterson-Falzone et al., 2009).

3) Speech therapy, which is a questioned method for improving velopharyngeal function. Over the years, many different techniques have been tried, but the results have often been disappointing (Peterson-Falzone et al., 2009; Sell & Grunwell, 2001). When the individual is able to achieve velopharyngeal closure in some cases, but the function is inconsistent and not used on all speech sounds, speech therapy is usually recommended. The focus of therapy is then to increase the person's awareness of when velopharyngeal closure is achieved and sometimes learn a new pattern for producing a specific sound that has been learned in the wrong way (most commonly the nasal production of /s/).

The treatment of articulation disorders is usually performed by an SLP or a teacher at school. Some advocate early stimulation via parents to encourage the child to produce anterior sounds in babbling, hoping thus to prevent later speech errors (Peterson-Falzone, 2009; Russell & Harding, 2001). Direct therapy can be used from the age of about 3 years, although not all children are inclined to cooperate at this age. As the child grows older, therapy can be more specifically targeted at the difficult speech sounds, with the emphasis on promoting new motor patterns in isolated speech sounds (articulatory therapy), or on whole words and efforts to increase awareness of phonemic borders in minimal pairs (phonological therapy). There are indications that frequent and focused training works best when it comes to changing articulatory patterns (Russell & Harding, 2001) and, to achieve the best possible speech outcome, "early and aggressive" management is advocated by Kuehn and Moller, among others (2000). However, the effectiveness of the different approaches has not yet been demonstrated. Although the person's communicative function is usually taken into consideration when the intervention is planned, the person's own view of the communicative situation is not assessed in a structured way at present (Sell, 2005).

Individuals' own satisfaction with treatment outcome

There appears to be a growing consensus that a person's own satisfaction with the treatment he or she has received is an important, if not the most important,

factor when health care is evaluated (e.g. Semb et al., 2005). The timing of the assessments of treatment satisfaction has varied. In a number of studies, researchers have approached young adults after their treatment has been terminated to obtain a final subjective evaluation of treatment outcome (e.g. Farzaneh et al., 2008; 2009; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Oosterkamp et al., 2007). Other studies have investigated satisfaction with outcome in children and adolescents among both the individuals with a cleft themselves and their parents (Broder et al., 1992; Hunt et al., 2006; Noor & Musa, 2007; Semb et al., 2005; Strauss et al., 1988). The majority of both individuals with a cleft and their parents report that they are satisfied with their speech outcome and satisfaction increases as the children grow older (Broder et al., 1992). It should be noted that a few remain dissatisfied with their speech into adulthood. However, dissatisfaction with appearance is more commonly observed and it has been found to correlate with factors related to quality of life (Marcusson et al., 2002; Oosterkamp et al., 2007; Sinko et al., 2005). Most of the research dealing with quality of life has focused on the associations with dissatisfaction with appearance and, at present, there is no proof that dissatisfaction with speech is associated with poorer quality of life.

Another question has been whether children or adolescents agree with their parents about how satisfied they are with treatment outcome. Broder and colleagues (1992) compared satisfaction with appearance and speech in 5- to 18-year-olds and found that the parents of females expressed more concern about their daughters' appearance than the parents of males, who were more concerned with their sons' speech. Another study of 13- to 18-year-olds (Strauss et al., 1988) found that 9% of the patients thought their main problem was speech, while 13% mentioned their appearance. When their parents were asked about their teenagers' main problems, the areas were reversed; 10% thought it was their speech and 6% their appearance. Richman (1983) found that social introversion was more related to concerns with facial appearance than speech during adolescence. It then seems plausible that teenagers are particularly critical of their own appearance and it is possible that their speech would become a concern in later years.

International Classification of Functioning, Disability and Health (ICF)

To broaden the perspective when describing health and health-related states, the ICF was introduced by the World Health Organization in 2001 (see Figure 1) as a sequel to the previous International Classification of Impairment, Disability and Handicap (ICIDH). The ICF attempts to integrate the different perspectives of health from biological, individual and social angles and complements the International Classification of Diseases (ICD). The ICF focuses on the impact

rather than the cause of a disorder and aspires to introduce a “common metric” for reporting the individual experience of disability. It is a classification system consisting of two parts, where part one relates to a person’s functioning and disability and part two to the contextual factors. The first part is classified in a more elaborate way and consists of two main components. The first component relates to body functions and structures and the second to activities and participation in different life areas. The second part consists of the two components environmental factors and personal factors. The environmental factors that interact with the components activity and participation are listed and can be qualified as barriers or facilitators. The personal factors are not classified, as they are expected to be so different in different cultural contexts, but they are mentioned, as they play an important role in the way people cope with impaired functioning and disability.

	Part 1: Functioning and Disability		Part 2: Contextual Factors	
Components	Body Functions and Structures	Activities and Participation	Environmental Factors	Personal Factors
Domains	Body Functions Body Structures	Life Areas (tasks, actions)	External influences on functioning and disability	Internal influences on functioning and disability

Figure 1. Overview of the ICF (WHO, 2001)

Participation in individuals born with a cleft

Previous research concerning individuals born with cleft lip and palate and other craniofacial anomalies has identified many additional threats to their quality of life, in addition to deviant speech, such as a different appearance, possible hearing loss, a tendency to regurgitate food down their nostrils and frequent hospitalisation (Bradbury, 2001; Kapp-Simon, 2005). Most research about self perception and quality of life has focused on dealing with a different appearance. Many of these studies have included more severe types of

craniofacial anomaly than clefts, making group comparisons difficult to interpret with regard to how much is valid for the cleft group (e.g. Kapp-Simon & McGuire, 1997; Krueckeberg et al., 1993; Pope & Ward, 1997; Topolski et al., 2005; Warschusky et al., 2002). However, as a group, individuals born with a cleft have been found to be more anxious and less “difficult” in their behaviour (Bradbury, 2001) and less active in social interaction (Slifer et al., 2004). Large surveys investigating various quality-of-life-related variables in adults born with a cleft have found a number of differences compared with individuals without a cleft; that fewer individuals with a cleft marry and that anxiety, depression and palpitations are twice as common (McWilliams & Paradise, 1973; Ramstad et al., 1995a; 1995b), that they rate themselves lower in the areas of life meaning and quality of life (Marcusson et al., 2001) and that some of them, especially women, are dissatisfied with the aesthetic outcome of their treatment (Sinko et al., 2005). Self-satisfaction with appearance has also been found to correlate with psychosocial adjustment at 10-15 years of age (Thomas et al., 1997). An early study by Richman (1978) studied the effects of facial disfigurement on teachers’ perception of how intelligent the children were. Teachers tended to rate cleft children with facial disfigurement less accurately; highly intelligent children were underestimated, whereas less intelligent children were overestimated. In another study, Richman (1997) studied speech in cleft palate patients along with facial ratings in relation to different types of behaviour at three ages and found that less severe speech problems were associated with behavioural inhibition at age 9 years, while greater facial disfigurement was associated with greater inhibition at 12 years. In overall terms, the findings have been somewhat contradictory, but no clear connection has been shown between a cleft and psychological problems (Bradbury, 2001; Clifford, 1983; Richman, 1983; Strauss & Broder, 1991).

Communicative Participation

Definition

The Participation component of the ICF is defined as “involvement in a life situation”. The construct Communicative Participation has been defined by Eadie et al. (2006) as “taking part in life situations where knowledge, information, ideas, or feelings are exchanged. It may take the form of speaking, listening, reading, writing, or nonverbal means of communication... and must involve a communicative exchange” (p. 309). So, the scope is broad and communicative participation can be studied and described in many different ways, from the individuals’ own attitude to their speech and communication, observational studies of their social interaction and investigations of how people in their environment react to their speech, to surveys of demographic data, such as their educational level, employment and marital status.

Environmental Factors – listeners' impressions

Since communication presupposes at least one other person participating in the communicative exchange, it is important to study the Environmental Factors in terms of the communication partners involved. There is also a common belief that our self-concept is shaped by the feedback we receive from people in our environment, particularly significant people in our closest family during our early years (e.g. Erikson, 1968). There have been a few investigations of how speech in individuals with a cleft palate is perceived by lay listeners. Berry et al. (1997) compared the personality characteristics attributed to speech samples of a group of children with a group of unaffected children and found no differences. The majority of the children in the cleft group were, however, assessed as having normal speech and no comparisons based on different speech status were carried out. This makes it difficult to infer whether deviant speech was perceived in a negative way, as individual differences are “drowned” in the group comparisons, which is typical of many group comparisons of individuals with clefts due to the large individual variations in speech outcome. The participants with speech deviations are rarely identified for separate analysis and the ones with pronounced speech disorders are often not included at all.

A study that did single out speakers with deviant speech was conducted by Blood and Hyman (1977). They investigated how hypernasal speech in children was perceived by their peers, using audio samples of four girls assessed as having normal, mild, moderate and severe hypernasality respectively. The samples were played to 120 children from kindergarten to second grade, who subsequently answered five questions about the person speaking, such as “Did you like the person talking?”. The researchers classified the answers as positive, negative, or neutral and found that listeners responded more negatively to the voice samples as hypernasality increased. The youngest children were not as negative to the moderately hypernasal voice, but all the children were negative to the severely hypernasal voice. The numbers were very small in this study, but there was an indication that hypernasality is perceived in a negative way. Research into other types of speech disorder has found similar tendencies. Lass et al. (1993) asked 13-year-olds with normal speech to listen to tape recordings of 16 children aged 6-11 years. Eight of the children had dysarthric speech and eight normal speech. The listeners then rated probable personality traits for each speaker on 22 scales containing polarised adjective pairs, such as “not smart-smart” and “mean-nice”. The children with dysarthria were rated less favourably on all 22 scales, compared with the children with normal speech, and the differences were statistically significant. The same design was used with eight children with voice disorder compared with eight children with normal speech (Lass et al., 1991). The children with voice disorders were perceived in a

significantly more negative way on twelve of the 22 scales. This indicates that children with deviant speech can be met with negative attitudes from their peers. However, in order to reflect a situation representative of their actual communicative participation, more holistic and naturalistic types of study design need to be employed, since listening to isolated audio recordings disregards the personal interaction between the children and people in their environment.

There has been some concern that SLPs that specialise in cleft palate speech have become “overtrained” in noting minor deviations in speech and are therefore not representative of the people a person meets in a natural environment. In an effort to create more naturalistic listener situations, untrained listeners were exposed to audio recordings of deviant speech of varying degrees. The speech ratings made by the untrained listeners roughly agreed with speech assessments made by specialised SLPs, although the untrained listeners did not always note minor signs of VPI (Brunnegård et al., 2009).

Personal Factors

The Personal Factors comprise features of the individual that are not specifically a part of their health state. They are made up of a person’s internal factors, such as gender, age, education, social background, coping style and other factors that influence the way disability is experienced by the individual (WHO, 2002). The Personal Factors are mentioned but not listed in the ICF, as they are considered to be dependent on the cultural context in which the person lives and are therefore difficult to qualify and compare between different cultural contexts. More recently, some researchers have attempted to identify and describe the personal qualities that enable some individuals to cope more effectively and be resilient in spite of disease and impaired functions (e.g. Bilboul et al., 2006; Feragen et al., 2009).

Holistic aspects of communication in quantitative research

As mentioned above, participation in itself is an holistic concept and communicative participation can be difficult to isolate from participation as a whole. However, efforts to decide what the consequences of a speech impairment may be have attracted some interest in the past. In the field of speech-language pathology, much of the research and clinical work focusing on the individuals’ communicative situation has been undertaken with people who stutter. The reason for this is probably that those professionals have regarded stuttering as involving more than the presence of outward manifestations of a speech disorder, since in some cases a negative attitude to speech and communication can impede the improvement and maintenance of fluency (Vanryckeghem, 1995). A negative attitude to communication and a tendency to

avoid activities that involve speech can be seen as an integral part of the stuttering syndrome and it needs to be addressed in therapy as well as in evaluations of treatment outcome. The development of these measures of the individual's own attitude to speech and communication has therefore been an ongoing process for some 40 years. In the 1960s, Lanyon (1967) and Erickson (1969) designed questionnaires for adults who stuttered and, in 1989, Gene Brutton designed a communication attitude test for schoolchildren (CAT) (Brutton & Dunham, 1989). The CAT is part of the Behavior Assessment Battery (BAB) and consists of 35 statements which investigate the child's speech-associated belief system, such as "I like the way I talk", "Some kids make fun of the way I talk", "I don't worry about talking on the phone", "It is hard for me to talk to strangers". The child classifies each statement as "true" or "false". Answers that indicate a negative attitude to speech and communication are given a score of one, so that a higher total score indicates a more negative communication attitude. The CAT has been used with children who stutter compared with children with normal speech and a significantly more negative communication attitude was found in the children who stuttered (DeNil & Brutton, 1991). They became more negative with increasing age, whereas the children who did not stutter became less negative after the age of 9 years. In 2000, a Swedish translation of the Communication Attitude Test, called the CAT-S, was made (Johannisson & Wennerfeldt, 2000). Reference data for children with normal speech have been obtained, resulting in a mean value of 6.05 in a group of 220 children aged 7-15 years. Two types of administration were investigated, a group setting and with individual assistance if required for the 10-year-olds. No statistical differences were found between the two types of administration, resulting in a mean value of 5.91 (SD=3.53) for the group setting and 5.11 (SD=2.64) for the individual setting (Johannisson et al., 2009).

The Speech Participation and Activity of Children (SPAA-C)(McLeod, 2004) is a questionnaire designed for children with any type of speech impairment according to the structure of the ICF, with the emphasis on the categories of Activity and Participation, Environmental and Personal Factors. It is directed not only at the individual child but also at significant others in the child's environment, such as parents, siblings, peers and teachers. The authors themselves note that asking a child's friends about their impression of the child's speech involves a risk of increasing their friends' awareness of the child's speech impairment in a negative way and should be used with caution.

Summary of holistic findings in quantitative research for individuals born with a cleft

The impact of a cleft on a person's quality of life has been addressed and, although some individuals appear to suffer from social inhibition and more

anxiety, there is no evident association between being born with a cleft and a poorer quality of life (Bradbury, 2001; Clifford, 1983; Richman, 1983; Strauss & Broder, 1991). Others have assumed that less successful treatment results from the viewpoint of the professionals involved in cleft care will have a negative impact on that person's quality of life, but no such associations have been demonstrated either. However, if the person him- or herself is dissatisfied with his/her treatment outcome in terms of appearance, there is an association with a poorer quality of life (Marcusson et al., 2002; Oosterkamp et al., 2007; Sinko et al., 2005). Moreover, if the person reports being teased, an association with poorer psycho-social functioning has been found (Hunt et al., 2006). What remains to be investigated is whether there is an association between speech status as assessed by speech-language pathologists or the person's own satisfaction with speech and any influence on quality of life or communicative participation (Table 1).

Another question has been whether individual satisfaction with speech agrees with speech assessments made by trained speech-language pathologists. The answer has been that they do not correlate for the adolescents that have been studied (Semb et al., 2005). This is similar to comparisons of satisfaction with appearance with professional or instrumental evaluations of treatment outcome, where no correlations have been found (Mani et al., 2010; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Semb et al., 2005; Sinko et al., 2005; Turner et al., 1997). The lack of agreement is an indication that there are other factors that influence how satisfied one feels with oneself and, if greater quality of life is the aim of treatment, a "perfect" result from the viewpoint of the specialist may not be the most important target.

In the studies mentioned above, scales, self-report questionnaires and terminology of different types have been used, which makes it difficult to know whether there are any differences between different treatment concepts. Moreover, as only associations have been found but no direction of any causal relationships, we cannot know whether dissatisfaction with appearance leads to poorer quality of life, or whether poorer quality of life leads to dissatisfaction, or whether both depend on another unknown third factor. Rather than having the researcher speculate about the factors that influence the different outcomes, it seems like a good option to ask the person involved him or herself.

Table 1. Overview of research on how different aspects of the cleft are related to each other; being born with a cleft, quality of life (QOL), appearance or speech assessed by professionals, the individuals' own satisfaction with their appearance or speech, reported teasing, psychosocial functioning and communicative participation. \nleftrightarrow = no association found, \downarrow = low level of, \leftrightarrow = association found, $\nleftrightarrow?$ = association not investigated

Cleft	\nleftrightarrow	QOL \downarrow
Appearance ass \downarrow	\nleftrightarrow	QOL \downarrow
Speech ass \downarrow	\nleftrightarrow	QOL \downarrow
Appearance ass \downarrow	\nleftrightarrow	appearance sat \downarrow
Speech ass \downarrow	\nleftrightarrow	speech sat \downarrow
Appearance sat \downarrow	\leftrightarrow	QOL \downarrow
Teasing	\leftrightarrow	psychosoc func \downarrow
Speech ass	$\nleftrightarrow?$	QOL
Speech ass	$\nleftrightarrow?$	comm part
Speech sat	$\nleftrightarrow?$	QOL
Speech sat	$\nleftrightarrow?$	comm part

Theoretical foundations in qualitative methods

Qualitative methodology is often used when human beings and their participation in society are the object of study. It was originally used primarily within the fields of sociology, anthropology and psychology and aims to investigate social phenomena from the viewpoint of the people in question themselves. It is used to identify new questions rather than answers and to understand human processes rather than explain them (Malterud, 2001). It presupposes that human beings act towards things on the basis of the meaning those things have for them, that the meaning arises from the social interaction with other people and is handled by the individual in an interpretative process.

In other words, the focus of interest is not the objective meaning of the studied phenomenon but the subjective experience of it. This approach can be seen as a basis for all qualitative methods. Individual interviews are often used to ask open questions to allow the persons concerned to describe their experiences themselves. The “grounded theory” method (Glaser & Strauss, 1967) has its roots in symbolic interactionism, focuses in particular on the interpretative processes people use to understand their reality and aims to generate theoretical frameworks which explain the collected data. Grounded theory can be seen as an intermediate form between quantitative and qualitative research, since it includes a phase of formulating and testing an emerging hypothesis, thereby blending the deductive methods typical of quantitative research and the inductive, or empirical, methods typical of qualitative research. The hypothesis is to be grounded in the data that are obtained during the interviews and must not influence the questions that are asked from the start. Grounded theory analysis consists of the systematic coding of data using constant comparisons. Raw data are coded as they are collected step by step and re-coded later on a more abstract level. The analysis should thus prompt theory development rather than the verification of pre-existing theories. The participants are selected strategically so that they represent different genders and degrees of disorder within the studied group, for example. Data collection and analysis proceed simultaneously until the answers no longer add new information, so-called theoretical saturation (Dellve et al., 2002).

The terminology for assessing the quality and relevance of qualitative studies varies among different researchers, but Malterud (2001), for example, advocates a set of standards by which to check studies, such as Reflexivity (the self-awareness with which the researcher conducts his or her research during the whole process), Preconceptions (previous knowledge and beliefs about the area under study) and Transferability (what some describe as being equivalent to the quantitative research concept of external validity; the opportunity to draw conclusions about the findings beyond the studied population).

Summary of findings in qualitative research for individuals born with a cleft

Not many qualitative studies that include people born with a cleft have been conducted so far. In 2003, Patel and Ross interviewed adults born with a cleft lip and palate in South Africa about their cleft experience. They described being generally satisfied with the treatment they had received and perceived their speech to be intelligible. They had, however, been greatly affected by other people’s negative attitudes about the cleft and had been teased during their childhood. Chetpakdechit and colleagues (2009) interviewed twelve young adults born with an isolated cleft palate and a bilateral complete cleft lip

and palate in Sweden and found a core category, Hoping to be like other people. Johansson and Ringsberg (2004) interviewed parents about their experience of giving birth to a child with a cleft. They described how it took some time to adapt to the situation after the child was born. The parents were satisfied with the care from the cleft palate team, but they were sometimes troubled by responses from other people in their environment, including health care personnel who had low levels of knowledge about clefts. Some parents expressed concern about the future outcome of their children's speech and appearance but did not perceive their child as handicapped.

More information about the way individuals born with a cleft palate describe their speech and communication themselves is needed in order to identify the most important areas for intervention. Moreover, specific descriptions from those who have, or have had a speech impairment beyond their pre-school years, as to how they perceive and deal with their communication is lacking.

Aims

The overall aim of this thesis was to explore how cleft-related speech impairments, whether temporary or permanent, influence the affected individual's life, in particular regarding communicative participation, from a long-term perspective.

The specific aims were to answer the following questions, also summarised in Appendix 1.

- How do 10-year-old children born with a cleft involving the palate respond to the Swedish translation of the Communication Attitude Test compared with children without a cleft? Are there differences with regard to different cleft types or genders? (Study I)
- How do the responses of the children correlate with their parents' answers to questions about their perception of their children's speech and communication? (Study I)
- How do the responses of the children and their parents correlate with speech assessments made by SLPs? (Study I)

- How do young adults rate their satisfaction with the outcome of speech, nose, lip and teeth? (Study II)
- How does the adults' satisfaction with speech correlate with speech assessments made by SLPs? (Study II)
- How do the adults' satisfaction with the outcome of speech, nose, lip and teeth correlate with how often they think and are asked questions about the cleft? (Study II)

- How do young adults describe their experience of growing up with a cleft and a cleft-related speech impairment? (Study III)

- How do young adults describe their experience of growing up with a cleft-related speech impairment and the way it was dealt with? (Study IV)

Methods

Participants

Children

In Study I, 54 10-year-olds participated. All cleft types involving the palate were included and all children who could understand and respond to the statements were invited to participate when they came for their routine assessment, which usually took place in the month of their tenth birthday. The 54 participants born with a cleft were compared with a previously documented reference group of 35 10-year-olds with normal speech (Johannisson et al., 2009).

Adults

Young adults who had acquired some distance to their more intense phase of cleft treatment were chosen. Thirty-five adults aged 22-32 years (mean age 25 years) participated in Study II. They were born with complete clefts; 25 with unilateral and 10 with bilateral clefts. Another thirteen adults aged 25-34 years participated in Studies III and IV. They were born with unilateral or bilateral complete clefts or an isolated cleft palate and had been assessed as having a speech impairment when they came for their last routine visit to the cleft clinic in their late teens.

Procedure in the quantitative methods

Data collection

The participants in Study I completed the CAT-S (the Swedish version of the Communication Attitude Test), which consists of 35 statements about their communication attitude. Their parents completed a short parent questionnaire regarding their own satisfaction with their child's speech, if other people commented on their child's speech and whether they were intelligible at home, at school and with strangers.

The participants in Study II were asked to indicate whether they thought their speech was normal or not and whether they wanted to correct their nose, lip, teeth, or speech. They were also asked to estimate how often they thought about their cleft and how often other people asked them questions about their speech, nose, or lip. The choices were "every day", "once a week", "once a month", "occasionally" and "never". They were also asked to rate their general

satisfaction with their speech, nose, lip and teeth on four visual analogue scales. The scales were 100 mm each and the far end to the left was labelled “poor”, while the far end to the right was labelled “excellent”. A mark in the middle (50 mm from each end) was labelled “so-so”.

Speech material

In Study I, all 54 participants were audio recorded at 10 years of age, 50 of them had recordings from 7 years of age and 45 from 5 years of age. In Study II, all 35 participants were audio recorded on their visit to the cleft clinic. The recordings were made in a standardised manner using high-quality equipment and standardised speech material consisting of standard sentences with high-pressure consonants, nasal consonants and combinations of nasals and plosives. The adult participants also read a short text, whereas the children named pictures in single words.

Perceptual assessments

In both studies, perceptual speech assessments were made blindly and independently by two experienced speech-language pathologists (SLPs). In Study I, a four-point ordinal scale was used for perceptual overall ratings of velopharyngeal function, articulation skill, intelligibility and general impression of speech. In Study II, a five-point scale was used for perceptual overall ratings of velopharyngeal function, hypernasality, hyponasality, weak-pressure consonants, nasal emission, velopharyngeal friction and compensatory articulation, while a three-point scale was used to rate general impression of speech.

Reliability of speech assessments

In both studies, exact agreement (“point-by-point”) was calculated: the number of identical assessments was divided by the total number of assessments made. To calculate intra-rater agreement, about 30% of the items were doubled and were therefore assessed twice by each SLP in Study I, while the corresponding figure was about 20% in Study II. To calculate inter-rater agreement, the two SLPs made individual assessments of the entire material first and the exact point-by-point agreement was calculated. They then compared their results. When their assessments differed, they listened again and discussed their results in order to reach consensus in a mutual assessment. In Study I, the total intra-rater agreement was 82% and 80% respectively. The total exact inter-rater agreement was 69%, ranging from 58% for velopharyngeal function to 76% for intelligibility. In Study II, the total inter-rater agreement was 78%. The two raters had a total intra-rater agreement of 89% and 87% respectively. As the

levels for inter-rater agreement were not entirely satisfactory, the consensus assessments were presented as speech results.

Statistical analyses

The data were entered into SPSS (Statistical Package for Social Sciences) for Windows (version 13). In Study I, a mixture of parametric and non-parametric statistics was used. Correlations were calculated with Spearman's correlation coefficient and comparisons between genders and cleft types were made with Mann-Whitney U tests. However, the CAT-S scores were compared using t-tests, since this is a standardised test and earlier studies have used parametric statistics. In Study II, non-parametric statistics were used throughout, based on the assumption that speech assessments constituted rank orders and normal distribution could not be assumed. All statistical calculations were two-tailed and significant results refer to a 5% level.

Procedure in the qualitative methods

Data collection

Data were collected in semi-structured individual interviews which lasted for 80-120 minutes and took place at the participant's home, workplace or the university, according to the participant's choice. All interviews were audio-taped and thematic questions, such as "Could you tell me what it was like to grow up with a cleft" and "How did you feel about your speech?" were asked, with follow-up questions. Additional themes that were pursued were whether they had talked about the cleft and their speech to other people, had been bullied, or felt that the cleft had an impact on their lives at the present time and what they would like to tell a young person born with a cleft, parents of a newborn child with a cleft, or the professionals who work with cleft care.

Analysis

As this study mainly followed the guidelines of Grounded Theory, data collection and analysis proceeded simultaneously. To enhance reflexivity, the first author wrote memos during the entire research process and shared them with the co-authors. The full interview transcriptions were read in full by all authors and subsequently discussed, along with emerging categories. The initial coding of the first four interviews was performed independently by the first two authors and the content of the codes agreed with regard to content. The quotations presented are intended to facilitate the reader's evaluation of the trustworthiness of the findings.

Results

Communication attitude in children (Study I)

The 10-year-old children born with a cleft had a significantly more negative communication attitude compared with a reference group of children without a cleft (Johannisson et al., 2009) ($t(84.324) = 4.566, p < .001$) and a larger range (Figure 2). There were no differences between genders or cleft types.

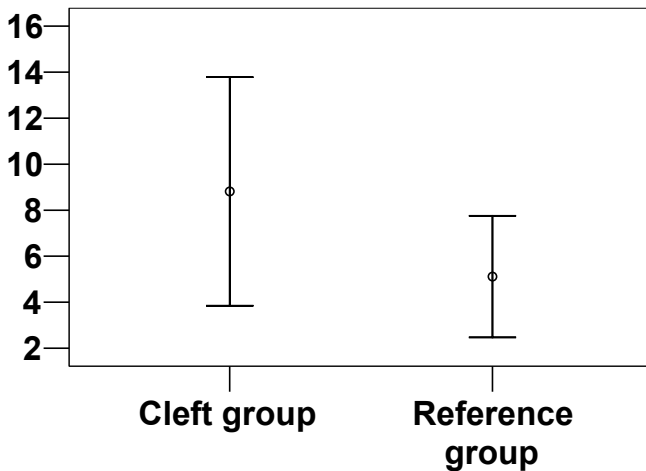


Figure 2. The mean CAT-S score +/- one standard deviation in the group with a cleft (n=54) and the reference group (n=35) (Johannisson et al., 2009).

The attitudes of children compared with the impressions of parents

All the answers from the parents were significantly correlated with the children's responses to the CAT-S (Table 2). The strongest association was with intelligibility with strangers and the lowest were intelligibility at home and being teased or not.

Table 2. Answers to the parent questions about environmental factors and their correlations with the CAT-S. The answers are graded so that, the higher the score, the more negative the communicative situation. Answers to the last question were categorised into no=0 or yes=1.

Parent question	Answers (%)				Correlation with CAT-S
	0	1	2	3	
Parent satisfaction	43	46	11	0	.448**
Comments from others	39	34	21	6	.474**
Intelligible at home	78	22	0	0	.334*
Intelligible at school	57	37	6	0	.541**
Intelligible with strangers	50	35	15	0	.649**
Teased (no/yes)	61	39	-	-	.358**

* $p < 0.05$, ** $p < 0.01$

Communication attitude compared with speech assessments

There were weak yet statistically significant correlations between the children's responses to the CAT-S and the speech variables assessed by SPLs, apart from velopharyngeal function at ages 5 and 7 years.

Satisfaction with treatment outcome in young adults (Study II)

None of the participants was more dissatisfied with his/her speech than "so-so" (Figure 3). Their satisfaction with speech was not statistically correlated with their thinking or being asked questions about the cleft.

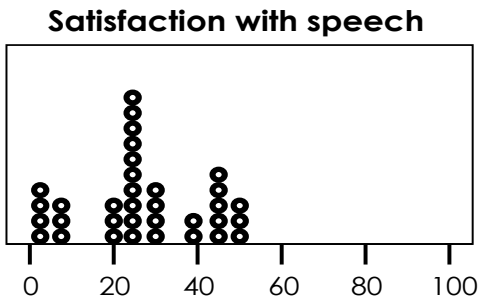


Figure 3. Satisfaction with speech rated on the VAS. 0=excellent, 50=“so-so” and 100=poor.

Young adults’ descriptions of their cleft experience (Study III)

The participants described how their experience of growing up with a cleft had influenced their lives and their descriptions were summarised by the core category of Making Sense of the Cleft. Categories under this headline are shown in Table 4.

Table 4. Making Sense of the Cleft. Overview of the core, main and subcategories.

Making Sense of the Cleft	
<p>Shaping one’s attitude to the cleft</p> <ul style="list-style-type: none"> • Forming an idea of the cleft • Choosing to be open about the cleft or not • Enduring hardships • Finding meaning in the cleft 	<p>Dealing with being different</p> <ul style="list-style-type: none"> • Orienting towards normality • Comparing oneself with others • Deciding when not to have more treatment

Young adults' descriptions of their speech and communication (Study IV)

The core category Taking Charge of Communication emerged from the analysis of the descriptions of how the participants perceived and handled their speech and communication. This described a gradual process of taking more active responsibility for speech and communication, made up of three categories with subcategories (Table 5).

Table 5. Taking Charge of Communication. Overview of the core, main and subcategories.

Taking Charge of Communication		
<p>Forming an idea of one's speech</p> <ul style="list-style-type: none"> • Making one's own assessment <ul style="list-style-type: none"> – Listening to recordings – Being attentive to one's own speech • Taking in other people's views <ul style="list-style-type: none"> – Considering professional assessments – Considering comments and reactions • Building conceptions of speech <ul style="list-style-type: none"> – Taking in how one's speech has changed – Reflecting on causes and effects 	<p>Learning about one's communication</p> <ul style="list-style-type: none"> • Monitoring the communicative situation <ul style="list-style-type: none"> – Understanding the listener's perspective – Understanding the demands of different speech situations • Knowing one's communicative "comfort zone" <ul style="list-style-type: none"> – Acknowledging one's own feelings – Understanding one's own responses 	<p>Taking responsibility for communication</p> <ul style="list-style-type: none"> • Changing the situation <ul style="list-style-type: none"> – Making decisions about speech intervention – Confronting speech • Accepting the situation <ul style="list-style-type: none"> – Focusing on things other than speech – Choosing to think constructively about one's own speech – Being open about one's speech disorder

Discussion

Attitudes to speech and communication in children born with a cleft

The 10-year-olds born with a cleft in Study I had a significantly more negative communication attitude compared with the reference children of the same age. They also had a wider range, which could be seen as typical for the cleft group, where many are well functioning and a few have problems (e.g. Richman, 1983). About two thirds had CAT-S scores within the range of the reference group and the rest had a more negative communication attitude. Study I also established significant correlations between the parents' answers about some environmental factors and CAT-S scores, as well as between speech results and CAT-S scores. In short, most children with high CAT-S scores had or had had a speech impairment, but quite a few children who had or had had a speech impairment had CAT-S scores within the normal range. Different children thus perceive and handle their speech impairments in different ways and some children would probably benefit from interventions directed at their communicative situation as a whole. The first step would be to include assessments of their attitude to their speech and communication at their routine visits to the cleft clinic. This is in agreement with the findings in research about psychosocial resilience, advocating that the individual experience of impairment should be assessed and form the basis of intervention (Feragen et al., 2009).

The CAT was developed for children who stutter, but some of the statements that could be assumed to be “stuttering specific”, such as “Sometimes words will stick in my mouth when I talk”, were classified as also being true for some of the children with typical speech development. This indicates that some of the vocabulary that we as SLPs consider specific to a particular diagnosis may represent a more general experience of speaking and the same questionnaire could therefore be used for different types of speech impairment. The overall CAT score, however, does not say anything specific about how a person functions communicatively, as both awareness of reduced intelligibility and reluctance to participate in communicative situations are combined into one total score. The CAT also contains some problematic statements such as the somewhat abstruse statement “The other kids wish they could talk like me”, which is considered to indicate a negative attitude to communication if responded to as “False”. Several children who completed the CAT-S were bewildered by that particular statement and one girl said “Perhaps someone small, who can't talk?”. Another statement was “I would

rather talk than write”, where the response “False” is also interpreted as indicating a negative communication attitude. Several children commented on this statement and said that they really liked both writing and talking and found it difficult to choose. This indicates that some of the statements are difficult to interpret as to whether they really have any bearing on a person’s communicative attitude or participation. A lasting impression from conducting Study I was that the information in the children’s spontaneous comments was at least as interesting as their total CAT-S score.

Attitudes to speech and communication in adults born with a cleft

There was no correlation between the persons’ own satisfaction with their speech and their answers to the questions about the influence of the cleft on their present life situation in Study II. This could be due to the fact that no participant in this study was more dissatisfied with their speech than “so-so” (the middle of the VAS), or that speech is considered less important than appearance. Previous studies have found that dissatisfaction with appearance is most common in adolescents born with a cleft (Broder et al., 1992; Semb et al., 2005) and that they think that speech is less important (Noor & Musa, 2007). Research has also indicated that appearance continues to be of greater importance than speech even in adulthood (Sinko et al., 2005). Other studies have, however, found that improving speech is deemed the most important part of treatment in cases where treatment has been offered through a “medical mission” (Sharp et al., 2008). Many people may be unaware of how they sound, as several participants in the interviews mentioned that it was primarily when they heard themselves on recordings that they became aware of the way they sounded and that this insight was painful. The lack of agreement between professional speech assessments and own satisfaction with speech that was also found in Study II could be another sign of this unawareness. However, this agrees with the lack of agreement observed between satisfaction with appearance and professional evaluations of it (Mani et al., 2010; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Semb et al., 2005; Sinko et al., 2005) and may simply be a reflection of the personal factors that make different people perceive and handle their impairments in different ways. In the interview study about communication (Study IV), one of the most important facilitating factors appeared to be the ability to see things from the viewpoint of the listener and to be open about the cleft and its effect on speech. Some of the participants were well aware of their speech impairment and let that knowledge guide them in the way they developed strategies for improving communication, such as telling their communication partner they were sometimes difficult to understand and encouraging the listener to ask them to repeat themselves if their speech was unclear. Again, speech impairments are handled differently by different

individuals and they have individual needs for intervention. Assessing the experienced communicative participation of the person born with a cleft, along with the assessment of speech status performed regularly at most cleft centres, would make an important contribution to the understanding of the person's communicative situation as a whole and provide important indications for intervention. There is a need for an instrument that measures communicative participation in children and adolescents born with a cleft, as has previously been identified (Sell, 2005).

Growing up with a cleft

The participants' descriptions contained different ways of making sense of the cleft and, although their stories differed from each other, they also had common features. The persons who were open about their cleft and their speech appeared to be the ones who were most positive about their cleft experience and found meaning in it. They were all positive about the care they had received and talked about how the members of the cleft team had worked to make the treatment outcome as good as possible and that this in itself was a positive experience that made them feel important. Their parents' importance as role models for how the cleft was dealt with was mentioned by several participants, but some of them thought they would do some things differently if they had children of their own with a cleft, as they had personal experience of growing up with a cleft that gave them a deeper insight into the situation. Another study interviewing young adults born with a cleft, however, identified the core category as Hoping to be like other people (Chetpakdeechit et al., 2009). The participants in the present interviews also talked about feeling different in an unwanted way, especially in their early teens, but they could also identify things that had made the cleft experience meaningful for them, some directly and effortlessly and others after some thought. The ability to see life events as meaningful and something to learn from has been identified as part of the Sense of Coherence concept developed by Antonovsky (1987) and it is thought to be an important component in promoting health. Constructing meaning in life is also an inherent human quality that is an underlying assumption in qualitative research (e.g. Dellve et al., 2002). Studies of self-concept in people born with a cleft have come to somewhat different conclusions; Persson and colleagues (2002) found similar or even higher self-concept in those born with a cleft compared with controls, whereas Leonard and colleagues (1991) found average or above average self-concept in children born with a cleft, but lower self-concept in teenage girls and higher self-concept in teenage boys. Directing attention at the people who feel exposed to negative comments and experience a lack of meaning in life is important and several participants in the interviews said that they would probably have needed to talk to a psychologist or therapist in their early teens, but thought that they would have refused to do so if someone had offered it to

them at the time. It is possible that, if contact with a psychologist was part of a routine, it would seem less intimidating and could benefit those born with a cleft during their difficult early teens.

Dealing with a cleft-related speech impairment

Some participants did not define their speech as “deviant” at the present stage but as something that had been a problem previously. Many of them talked about the great improvement that had occurred since childhood and that they were now largely satisfied with their speech. The relatively few individuals who had impaired articulation seemed to be more troubled by their speech and felt that they should have worked harder with their speech training. If our focus is on the impairment level, a speech impairment should be treated with the “early and aggressive” management approach advocated by some SLPs (Kuehn & Moller, 2000). However, some children and adolescents are reluctant to participate in frequent speech training, especially when progress is slow, and finding a balance between letting a child decide about intervention on the one hand and trying to encourage participation in therapy on the other is sometimes a difficult task for the SLP. Given the differences between professional assessments of speech status made by SLPs and the individuals’ own satisfaction with speech (as well as the lack of agreement between the professionals themselves, further discussed below), it is important that we do not continue treating speech disorders that do not constitute a problem for the affected individual in a long-term perspective. Communication is a complex phenomenon that includes more than just speech quality and sometimes a person’s communicative participation may be best improved by types of intervention other than speech training. The analysis of the interview study demonstrated that by learning about their communication and being attentive to their communication partners’ response, the participants could take responsibility for the communicative situation in different ways, and in this way their speech impairment did not result in restricted participation.

A speech impairment may involve a risk of being met with prejudice, at least when it comes to first impressions (Blood & Hyman, 1977; Lass et al, 1991; 1993). Providing information about the cleft and cleft-related speech disorders to people who meet children with clefts could be one way of overcoming misconceptions and could provide a young child with a model of how to deal with curiosity and comments from people in their environment. Since teasing has been found to influence a person’s psycho-social functioning (Hunt et al., 2006) and it has been established that cleft-related teasing is quite common (Noar, 1991; Noor & Musa, 2007; Semb et al., 2005; Turner et al., 1997), it is important to be attentive to such tendencies in a child’s environment and, if needed, help the child with strategies to deal with unwanted comments and questions about the cleft.

Methods employed in the studies

A mix of quantitative and qualitative methods was chosen to explore how the individuals themselves perceive and describe their speech and communication. In Study I, children's attitudes to communication were explored via a self-report instrument originally developed for children who stutter, the CAT, discussed earlier. A complicating factor in Study I was the low level of inter-rater agreement in the assessments of some speech variables. As low reliability in speech assessments was a known obstacle, a slightly reduced scale was used in Study I (a 4-point scale as opposed to the 5-point scale more commonly used in Sweden) in the hope that this would increase the reliability of the ratings (Sell, 2005). The 4-point scales were used for both the overall impact on speech (intelligibility and overall impression of speech) and the typical cleft-related speech impairments (velopharyngeal function and articulation disorders). However, the agreement was disappointingly low, especially in assessing velopharyngeal function (58%). According to the recommendations to use an external judge, who is not familiar with the individuals who are being assessed (Whitehill, 2002), an external judge from another Swedish cleft centre performed the assessments, in addition to a judge from the centre in Gothenburg. Although they were both experienced in assessing speech typical of individuals born with a cleft palate, they had not collaborated on assessment tasks before and it is possible that they were therefore not as "calibrated" as one might have wished. Structured mutual training before the assessment task could have improved the reliability, as found in a recent study that investigated the immediate and maintained effects of joint listening and subsequent discussion to reach consensus which resulted in good stability of judgements (Sell et al., 2009). Since the problem of low reliability in perceptual speech assessment was well known, the judges in Study I also performed consensus assessments and these were used as speech results. This was considered to provide some justification for drawing conclusions from the speech assessments, even though the inter-rater agreement was poor.

As a group, the children born with a cleft palate had a significantly more negative attitude to communication compared with reference data, but there was large variability and the study design provided no explanation of why not all children with a speech impairment had developed a negative communication attitude. Significant positive correlations with the majority of the speech variables and the total score on the CAT-S were found, but the correlations were weak and difficult to interpret when it came to how relevant they were in each individual case. Altman (1991) suggests the calculation of $100r^2$ to estimate how much of the variability in the data is explained by the

correlation coefficient and, when this method was applied to Study I, it follows that, although the correlations are statistically significant, they only explained about 20-40% of the variance. Other factors, then, must influence the result and, to find these unknown factors, other types of method appeared to be needed.

Qualitative methods are advocated when the research area is a person's own experience of a certain phenomenon in its natural context and the meaning it has for the individual (Malterud, 2001). When the persons themselves talk about their situation and the meaning different phenomena have for them, a more "in-depth" understanding is reached. Qualitative findings do not allow for comparisons between different cohorts and have little value when comparing different treatment protocols. However, the quantitative research addressing satisfaction with treatment and general well-being has used such different methodology and different terminology that it is almost impossible to compare treatment outcome and patient satisfaction between different treatment protocols as well. One of the advantages of quantitative methods is that they allow for comparisons between different populations, but, to make this possible, basic terminology and instruments must be agreed on.

What is the task of the cleft team?

It has been argued that the person's own satisfaction with his/her treatment outcome is an important, if not the most important, goal for cleft care (e.g. Semb et al., 2005). However, it seems that a person's satisfaction is associated more closely with personal and environmental factors than with the size of their deviation from the norm in professional assessments. So what is the task of the cleft team? Most people would agree that the cleft must be repaired with the best possible aesthetic result, and the teeth, hearing and speech should be as good as possible. Is high satisfaction the ultimate goal of treatment even if it says nothing about the result from a more objective assessment (by both lay people and professionals)? The lack of agreement between professional assessments and the individuals' own satisfaction with treatment results has been established in previous research (Mani et al., 2010; Meyer-Marcotty & Stellzig-Eisenhauer, 2009; Semb et al., 2005; Sinko et al., 2005; Turner et al., 1997). Sinko and colleagues (2005) also found that the individuals who desired further treatment had poorer quality of life. One important conclusion that can be drawn from this is the importance of always involving the affected individuals themselves in decisions about treatment. The concept of *burden of care* is given increasing focus (Semb et al., 2005), as it is thought to be of importance to a young person's quality of life and the impact of having to go through treatment that one is not personally motivated to undertake could be as negative as living with deviant speech or appearance. When asked what they would like to tell the

people who work with cleft care, the participants in the interviews had very few concrete suggestions for improvement. Some of them mentioned the discomfort of being “study material” during doctors’ rounds at the hospital, particularly when they were in their teens, but otherwise they were very satisfied with their care. Some of them specifically mentioned the positive feeling of having several professionals “wishing them well” and how it made them feel important and reassured that the cleft team members were focusing on their treatment outcome to make it as good as possible.

School has the task of providing an arena in which the child can be confirmed and develop as a social being, in addition to acquiring sufficient knowledge. Children spend much of their time at school and comparably little time in hospital. The experience of being bullied and teased was mentioned as the most negative thing about growing up with a cleft by the participants in the interviews and this supports previous research about the importance of social acceptance (Allison et al., 2009; Brunstein Klomek et al., 2007; Hunt et al., 2006). Providing an environment in which the children can feel accepted and appreciated by their peers is primarily the task of the staff in schools, but it may also be an important task for the members of the cleft teams to draw the staff’s attention to the risk of being bullied and teased for those born with a cleft.

What is the task of the speech-language pathologist in cleft care?

The previous focus on speech outcome needs to be complemented with attention to the communicative situation as a whole. Some children who at the present time had good speech results still had a negative communication attitude. Speech-language pathologists working with cleft care need to expand their focus of research to include the participation component of the ICF. In addition, more research on the evaluation of treatment outcome is needed and it is important not to continue treatment that does not realise its goals. Prolonged treatment that does not have any effect can be negative for the person doing the training. It is important to involve the older child or adolescent in decisions about treatment. Most SLPs have experienced the situation in which the parents want treatment and the child resists it. Learning more about when and how to treat speech is an important task for SLPs and must be targeted in future research.

Limitations

The CAT, as well as the CAT-S, includes statements that are difficult to interpret, e.g., “The other kids wish they could talk like me”, which is discussed above. Based on tests of internal consistency in which three statements (“The other kids wish they could talk like me”, “My friends don’t talk as well as I do” and “I talk better with a friend”) were found not to correlate with the total CAT score (Brutten & Dunham, 1989), the CAT has also been issued in a version with 32 statements, as well as a version with 33 statements. This makes it difficult to compare the results of the present study with those from other studies. The results from the study of satisfaction with speech and how it is related to the impact of the cleft as a whole are incomplete, as no participants were more dissatisfied with their speech than “so-so”. For this reason, it is not possible to know if more dissatisfied individuals would have had an influence on the overall impact of the cleft. Another complicating factor in the quantitative studies was the low level of inter-rater agreement in the speech assessments. This problem is not new, but it limits the opportunity to draw well-founded conclusions from the results. The findings of the interview studies are based on a few participants and cannot be generalised to apply to the whole cleft population. The sampling procedure was complicated by the fact that not many young adults had remaining speech impairments when they were adolescents. No one had pronounced difficulties with intelligibility and it would have been interesting to know whether this would have had a different impact on their lives. Moreover, the fact that the interviews were conducted by an SLP may have made the participants less inclined to disclose negative experiences of speech-language pathology.

Summary and clinical implications

This thesis has investigated the affected individuals’ attitude to speech and communication. One of its conclusions is that a wider focus on communication needs to be adopted and that the individual’s own attitude to communication in a wider sense needs to be assessed regularly, as speech impairments are handled differently by different individuals. In addition, more evidence of treatment outcome is needed to avoid time- and effort-consuming interventions that do not have the desired effect. Cleft team members need to be aware of their influence on the individual’s will to have more treatment and should be careful when suggesting interventions. Including the experiences and viewpoints of the affected individuals themselves is an important way to develop current methods.

Future research

Interviewing children and adolescents with a cleft-related speech impairment is an important next step in obtaining a better understanding of the way speech impairments are perceived and dealt with at younger ages. Recently, children with speech impairments with other origins were interviewed (McCormack et al., 2010) and their perspective provides important information about their communicative experience. It is also important to interview individuals with more pronounced speech impairments, such as glottal articulation. Individuals with profound speech impairments are often omitted in research, as they are thought to be problematic to interview due to their reduced intelligibility. It is, however, probable that they have experiences that are quite different from those of individuals with mild or moderate impairments. More studies of evidence of treatment effect to avoid time-consuming treatment that does not have the desired effect need to be conducted. In order to do this reliably, improved methods for assessing speech, in particular signs of velopharyngeal impairment, need to be developed. Another important area is to develop new self-report instruments for assessing communicative participation and satisfaction with speech based on the affected persons' own experiences of dealing with their speech impairments.

Acknowledgements

I would like to express my sincere gratitude to everyone who has contributed to this thesis and first and foremost to all the people born with a cleft who so generously gave their time and effort to answer my sometimes probing questions. In particular, I also want to thank

Anette Lohmander, my supervisor and co-author, for generously sharing your knowledge and always being inspiring and enthusiastic

Annika Dahlgren Sandberg, my co-supervisor and co-author, for encouragement and generous help with statistical analyses

Katja Laakso and Karin C Ringsberg, my co-authors in the qualitative papers, for your generous and insightful contributions to the analyses of the extensive interview material

Anna Elander, my co-author, for initiating Study II and generously handing over some of the data to me

Christina Persson and Karin Brunnegård, for kindly performing the tiresome perceptual speech assessments

Anna-Karin Norrman, for kind and never-ending help with practical matters

Friends and colleagues at the Division of Speech-Language Pathology at University of Gothenburg

Friends and colleagues at the Department of Logopedics at Sahlgrenska University Hospital

Members of the cleft palate team at Sahlgrenska University Hospital and, finally, family and friends for love and support through the years.

The studies were conducted with financial support via grants from the state under the ALF agreement, the Swedish Council for Working Life and Social Research and the Mayflower Foundation.

References

- Allison, S., Roeger, L., & Reinfeld-Kirkman, N. (2009). Does school bullying affect adult health? Population survey of health-related quality of life and past victimization. *The Australian and New Zealand Journal of Psychiatry*, 43, 1163-1170.
- Altman, D. G. (1991). *Practical Statistics for Medical Research*. London: Chapman & Hall.
- Antonovsky, A. (1987). *Unraveling the mystery of health: how people manage stress and stay well*. San Francisco, Calif., Jossey-Bass.
- Becker, M., Svensson, H., Sarnas, K. V., & Jacobsson, S. (2000). VonLangenbeck or Wardill procedures for primary palatal repair in patients with isolated cleft palate--speech results. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 34, 27-32.
- Berry, L. A., Witt, P. D., Marsh, J. L., Pilgram, T. K., & Eder, R. A. (1997). Personality attributions based on speech samples of children with repaired cleft palates. *Cleft Palate-Craniofacial Journal*, 34, 385-389.
- Bilboul, M. J., Pope, A. W., & Snyder, H. T. (2006). Adolescents with craniofacial anomalies: psychosocial adjustment as a function of self-concept. *Cleft Palate-Craniofacial Journal*, 43, 392-400.
- Blood, G. W., & Hyman, M. (1977). Children's perception of nasal resonance. *Journal of Speech and Hearing Disorders*, 42, 446-448.
- Bradbury, E. (2001). Growing up with a cleft: the impact on the child. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of Cleft Lip and Palate* (pp. 365-378). London: Whurr Publishers.
- Broder, H. L., Smith, F. B., & Strauss, R. P. (1992). Habilitation of patients with clefts: parent and child ratings of satisfaction with appearance and speech. *Cleft Palate-Craniofacial Journal*, 29, 262-267.
- Brunnegard, K, Lohmander, A., & van Doorn, J. (2009). Untrained listeners' ratings of speech disorders in a group with cleft palate: a comparison with speech and language pathologists' ratings. *International Journal of Language and Communication Disorders*, 44, 656-674.
- Brunstein Klomek, A., Marrocco, F., Kleinman, M., Schonfeld, I. S., & Gould M. S. (2007). Bullying, depression, and suicidality in adolescents. *Journal of the American Academy of Child and Adolescent Psychiatry*, 46, 40-49.
- Brutten, G. J., & Dunham, S. L. (1989). The communication attitude test. A normative study of grade school children. *Journal of Fluency Disorders*, 14, 371-377.

- Cassel, M. D., & Elkadi, H. (1995). Anatomy and physiology of the palate and velopharyngeal structures. In R. J. Shprintzen & J. Bardach (Eds.) *Cleft palate speech management. A multidisciplinary approach* (pp. 45-62). StLouis, MO: Mosby.
- Clifford, E. (1983). Why are they so normal? *Cleft Palate Journal*, 20, 83-84.
- Chapman, K. L., Hardin-Jones, M., & Halter, K. A. (2003). The relationship between early speech and later speech and language performance for children with cleft lip and palate. *Clinical Linguistics & Phonetics*, 17, 173-197.
- Chetpakdechit, W., Hallberg, U., Hagberg, C., & Mohlin, B. (2009). Social life aspects of young adults with cleft lip and palate: grounded theory approach. *Acta Odontologica Scandinavica*, 67, 122-128.
- Dellve L, Henning Abrahamsson K, Trulsson U, Hallberg LRM. (2002). Grounded theory in public health research. In R. L. M. Hallberg (Ed). *Qualitative Methods in Public Health Research: Theoretical Foundations and Practical Examples* (pp. 137-173). Lund, Sweden: Studentlitteratur.
- DeNil, G. J., & Brutten, G. F. (1991). Speech-associated attitudes of stuttering and nonstuttering children. *Journal of Speech and Hearing Research*, 34, 60-66.
- Dotevall, H., Lohmander-Agerskov, A., Ejnell, H., & Bake, B. (2002). Perceptual evaluation of speech and velopharyngeal function in children with and without cleft palate and the relationship to nasal airflow patterns. *Cleft Palate-Craniofacial Journal*, 39, 409-424.
- Eadie, T. L., Yorkston, K. M., Klasner, E. R., Dudgeon, B. J., Deitz, J. C., Baylor, C. R., et al. (2006). Measuring communicative participation: a review of self-report instruments in speech-language pathology. *American Journal of Speech-Language Pathology*, 15, 307-320.
- Engström, K., Fritzell, B., & Johansson, B. (1970). A study of speech improvement following palatopharyngeal flap surgery. *Cleft Palate Journal*, 7, 419-431.
- Erickson, R. L. (1969). Assessing communication attitudes among stutterers. *Journal of Speech and Hearing Research*, 12, 711-724.
- Erikson, E. H. (1968). *Identity, Youth and Crisis*. New York: W. W. Norton.
- Farzaneh, F., Becker, M., Peterson, A. M., & Svensson, H. (2008). Speech results in adult Swedish patients born with unilateral complete cleft lip and palate. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 42, 7-13.
- Farzaneh, F., Becker, M., Peterson, A. M., & Svensson, H. (2009). Speech results in adult Swedish patients born with bilateral complete cleft lip and palate. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 43, 207-213.

- Feragen, K. B., Borge, A. I., & Rumsey, N. (2009). Social experience in 10-year-old children born with a cleft: exploring psychosocial resilience. *Cleft Palate-Craniofacial Journal*, 46, 65-74.
- Friede H. (2009). Two-stage Palate Repair. In: J. E. Losee & R. E. Kirschner (Eds). *Comprehensive Cleft Care* (pp. 413-429). New York: McGraw Hill.
- Glaser, B. G., & Strauss, A. L. (1967). *The Discovery of Grounded Theory: Strategies for Qualitative Research*. New York: Aldine de Gruyter.
- Grunwell, P., & Sell, D. A. (2001). Speech and cleft palate/velopharyngeal anomalies. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of Cleft Lip and Palate* (pp. 68-86). London: Whurr Publishers.
- Grunwell, P., Brondsted, K., Henningsson, G., Jansonius, K., Karling, J., Meijer, M., et al. (2000). A six-centre international study of the outcome of treatment in patients with clefts of the lip and palate: the results of a cross-linguistic investigation of cleft palate speech. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 34, 219-229.
- Hagberg, C., Larson, O., & Milerad, J. (1998). Incidence of cleft lip and palate and risks of additional malformations. *Cleft Palate-Craniofacial Journal*, 35, 40-45.
- Henningsson, G., Kuehn, D. P., Sell, D., Sweeney, T., Trost-Cardamone, J. E., & Whitehill, T. L. (2008). Universal parameters for reporting speech outcomes in individuals with cleft palate. *Cleft Palate-Craniofacial Journal*, 45, 1-17.
- Hunt, O., Burden, D., Hepper, P., Stevenson, M., & Johnston, C. (2006). Self-reports of psychosocial functioning among children and young adults with cleft lip and palate. *Cleft Palate-Craniofacial Journal*, 43, 598-605.
- Johannisson, B., & Wennerfeldt, S. (2000). *Intensiv stamningsterapi för barn. En jämförande och deskriptiv studie av stamning hos barn som genomgått, respektive sökt till terapin*. Unpublished master's thesis in Swedish. University of Gothenburg, Gothenburg, Sweden.
- Johannisson, T. B., Wennerfeldt, S., Havstam, C., Naeslund, M., Jacobson, K., & Lohmander, A. (2009). The Communication Attitude Test (CAT-S): normative values for 220 Swedish children. *International Journal of Language and Communication Disorders*, 44, 13-25.
- Johansson, B., & Ringsberg, K. C. (2004). Parents' experiences of having a child with cleft lip and palate. *Journal of Advanced Nursing*, 47, 165-173.
- Jones, C. E., Chapman, K. L., & Hardin-Jones, M. A. (2003). Speech development of children with cleft palate before and after palatal surgery. *Cleft Palate-Craniofacial Journal*, 40, 19-31.

- Kapp-Simon, K. A., & McGuire, D. E. (1997). Observed social interaction patterns in adolescents with and without craniofacial conditions. *Cleft Palate-Craniofacial Journal*, 34, 380-384.
- Kapp-Simon, K. A., McGuire, D. E., Long, B. C., & Simon, D. J. (2005). Addressing quality of life issues in adolescents: social skills interventions. *Cleft Palate-Craniofacial Journal*, 42, 45-50.
- Karnell, M. P., & Van Demark, D. R. (1986). Longitudinal speech performance in patients with cleft palate: comparisons based on secondary management. *Cleft Palate Journal*, 23, 278-288.
- Krueckeberg, S. M., Kapp-Simon, K. A., & Ribordy, S. C. (1993). Social skills of preschoolers with and without craniofacial anomalies. *Cleft Palate-Craniofacial Journal*, 30, 475-481.
- Kuehn, D. P., & Moller, K. T. (2000). Speech and language issues in the cleft palate population: The state of the art. *Cleft Palate-Craniofacial Journal*, 37, 348.
- Lanyon, R. I. (1967). The measurement of stuttering severity. *Journal of Speech and Hearing Research*, 10, 836-843.
- Lass, N. J., Ruscello, D. M., Harkins, K. E., & Blankenship, B. L. (1993). A comparative study of adolescents' perceptions of normal-speaking and dysarthric children. *Journal of Communication Disorders*, 26, 3-12.
- Lass, N. J., Ruscello, D. M., Stout, L. L., & Hoffmann, F. M. (1991). Peer perceptions of normal and voice-disordered children. *Folia Phoniatrica*, 43, 29-35.
- Leonard, B. J., Brust, J. D., Abrahams, G., & Sielaff, B. (1991). Self-concept of children and adolescents with cleft lip and/or palate. *Cleft Palate-Craniofacial Journal*, 28, 347-353.
- Lilja, J., Friede, H., & Johanson, J. (1996). Changing philosophy of surgery of the cleft lip and palate in Göteborg, Sweden. In S. Berkowitz (Ed). *Cleft Lip and Palate. Perspectives on Management. Vol. II* (pp. 155-170). San Diego: Singular Publishing.
- Lohmander, A., & Olsson, M. (2004). Methodology for perceptual assessment of speech in patients with cleft palate: a critical review of the literature. *Cleft Palate-Craniofacial Journal*, 41, 64-70.
- Lohmander, A., Friede, H., Elander, A., Persson, C., & Lilja, J. (2006). Speech development in patients with unilateral cleft lip and palate treated with different delays in closure of the hard palate after early velar repair: a longitudinal perspective. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 40, 267-274.
- Lohmander, A., & Persson, C. (2008). A longitudinal study of speech production in Swedish children with unilateral cleft lip and palate and two-stage palatal repair. *Cleft Palate-Craniofacial Journal*, 45, 32-41.
- Lohmander, A., Willadsen, E., Persson, C., Henningsson, G., Bowden, M., & Hutters, B. (2009). Methodology for speech assessment in the

- Scandcleft project--an international randomized clinical trial on palatal surgery: experiences from a pilot study. *Cleft Palate-Craniofacial Journal*, 46, 347-362.
- Lohmander-Agerskov, A. (1996). *Speech in Children with Cleft Lip and Palate Treated with Delayed Closure of the Hard Palate*. Unpublished doctoral dissertation, University of Gothenburg, Gothenburg, Sweden.
- Lohmander-Agerskov, A., Soderpalm, E., Friede, H., Persson, E. C., & Lilja, J. (1994). Pre-speech in children with cleft lip and palate or cleft palate only: phonetic analysis related to morphologic and functional factors. *Cleft Palate-Craniofacial Journal*, 31, 271-279.
- Malterud, K. (2001). The art and science of clinical knowledge: evidence beyond measures and numbers. *Lancet*, 358, 397-400.
- Mani, M., Semb, G., & Andlin-Sobocki, A. (2010). Nasolabial appearance in adults with repaired unilateral cleft lip and palate: relation between professional and lay rating and patients' satisfaction. *Journal of Plastic Surgery and Hand Surgery*, in press.
- Marcusson, A., Akerlind, I., & Paulin, G. (2001). Quality of life in adults with repaired complete cleft lip and palate. *Cleft Palate-Craniofacial Journal*, 38, 379-385.
- Marcusson, A., Paulin, G., & Ostrup, L. (2002). Facial appearance in adults who had cleft lip and palate treated in childhood. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 36, 16-23.
- Mars, M. (2001). Facial growth. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of cleft lip and palate* (pp. 44-67). London: Whurr Publishers.
- McCormack, J., McLeod, S., McAllister, L., & Harrison, L. J. (2010). My speech problem, your listening problem, and my frustration: The experience of living with childhood speech impairment. *Language, Speech, and Hearing Services in Schools*, in press.
- McLeod, S. (2004). Speech pathologists' application of the ICF to children with speech impairment. *Advances in Speech-Language Pathology*, 6, 75-81.
- McWilliams, B. J., & Paradise, L. P. (1973). Educational, occupational, and marital status of cleft palate adults. *Cleft Palate Journal*, 10, 223-229.
- Mercer, N. S. G., & Pigott, R. W. (2001). Assessment and surgical management of velopharyngeal dysfunction. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of Cleft Lip and Palate* (pp. 258-285). London: Whurr Publishers.
- Meyer-Marcotty, P., & Stellzig-Eisenhauer, A. Dentofacial self-perception and social perception of adults with unilateral cleft lip and palate. (2009). *Journal of Orofacial Orthopedics*, 70, 224-236.

- Mossey, P. (2007). Epidemiology underpinning research in the aetiology of orofacial clefts. *Orthodontics and Craniofacial Research*, 10, 114-120.
- Noar, J. H. (1991). Questionnaire survey of attitudes and concerns of patients with cleft lip and palate and their parents. *Cleft Palate-Craniofacial Journal*, 28, 279-284.
- Noor, S. N., & Musa, S. (2007). Assessment of patients' level of satisfaction with cleft treatment using the Cleft Evaluation Profile. *Cleft Palate-Craniofacial Journal*, 44, 292-303.
- Oosterkamp, B. C., Dijkstra, P. U., Remmelink, H. J., van Oort, R. P., Goorhuis-Brouwer, S. M., Sandham, A., et al. (2007). Satisfaction with treatment outcome in bilateral cleft lip and palate patients. *International Journal of Oral and Maxillofacial Surgery*, 36, 890-895.
- Park, S., Saso, Y., Ito, O., Tokioka, K., Takato, T., Kato, K., et al. (2000). The outcome of long-term follow-up after palatoplasty. *Plastic and Reconstructive Surgery*, 105, 12-17.
- Patel, Z., & Ross, E. (2003). Reflections on the cleft experience by South African adults: use of qualitative methodology. *Cleft Palate-Craniofacial Journal*, 40, 471-480.
- Persson, C. (2004). *Speech and Language in Patients with an Isolated Cleft Palate and/or 22q11 Deletion Syndrome*. Unpublished doctoral dissertation, University of Gothenburg, Sweden.
- Persson, M., Aniansson, G., Becker, M., & Svensson, H. (2002). Self-concept and introversion in adolescents with cleft lip and palate. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 36, 24-27.
- Peterson-Falzone, S. J., Hardin-Jones, M. A., & Karnell, M. P. (2009). *Cleft Palate Speech*. St Louis, MO: Mosby.
- Pope, A. W., & Ward, J. (1997). Self-perceived facial appearance and psychosocial adjustment in preadolescents with craniofacial anomalies. *Cleft Palate-Craniofacial Journal*, 34, 396-401.
- Pulkkinen, J., Haapanen, M. L., Paaso, M., Laitinen, J., & Ranta, R. (2001). Velopharyngeal function from the age of three to eight years in cleft palate patients. *Folia Phoniatica et Logopaedica*, 53, 93-98.
- Ramstad, T., Ottem, E., & Shaw, W. C. (1995a). Psychosocial adjustment in Norwegian adults who had undergone standardised treatment of complete cleft lip and palate. I. Education, employment and marriage. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 29, 251-257.
- Ramstad, T., Ottem, E., & Shaw, W. C. (1995b). Psychosocial adjustment in Norwegian adults who had undergone standardised treatment of complete cleft lip and palate. II. Self-reported problems and concerns

- with appearance. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, 29, 329-336.
- Richman, L. C. (1978). The effects of facial disfigurement on teachers' perception of ability in cleft palate children. *Cleft Palate Journal*, 15, 155-160.
- Richman, L. C. (1983). Self-reported social, speech, and facial concerns and personality adjustment of adolescents with cleft lip and palate. *Cleft Palate Journal*, 20, 108-112.
- Richman, L. C. (1997). Facial and speech relationships to behavior of children with clefts across three age levels. *Cleft Palate-Craniofacial Journal*, 34, 390-395.
- Russell, V. J., & Harding, A. (2001). Speech development and early intervention. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of Cleft Lip and Palate* (pp. 191-209). London: Whurr Publishers.
- Scherer, N. J., Williams, A. L., & Proctor-Williams, K. (2008). Early and later vocalization skills in children with and without cleft palate. *International Journal of Pediatric Otorhinolaryngology*, 72, 827-840.
- Sell, D. (2005). Issues in perceptual speech analysis in cleft palate and related disorders: a review. *International Journal of Language and Communication Disorders*, 40, 103-121.
- Sell, D. A., & Grunwell, P. (2001). Speech assessment and therapy. In A. C. H. Watson, D. A. Sell, & P. Grunwell (Eds). *Management of Cleft Lip and Palate* (pp. 227-257). London: Whurr Publishers.
- Sell, D., Grunwell, P., Mildinhal, S., Murphy, T., Cornish, T. A., Bearn, D., et al. (2001). Cleft lip and palate care in the United Kingdom--the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. *Cleft Palate-Craniofacial Journal*, 38, 30-37.
- Sell, D., John, A., Harding-Bell, A., Sweeney, T., Hegarty, F., & Freeman, J. (2009). Cleft audit protocol for speech (CAPS-A): a comprehensive training package for speech analysis. *International Journal of Language and Communication Disorders*, 44, 529-548.
- Semb, G., Brattstrom, V., Molsted, K., Prah-Andersen, B., Zuurbier, P., Rumsey, N., et al. (2005). The Eurocleft study: intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 4: relationship among treatment outcome, patient/parent satisfaction, and the burden of care. *Cleft Palate-Craniofacial Journal*, 42, 83-92.
- Sharp, H. M., Canady, J. W., Ligot, F. A., Hague, R. A., & Gutierrez, J. (2008). Caregiver and patient reported outcomes after repair of cleft lip and/or palate in the Philippines. *Cleft Palate-Craniofacial Journal*, 45, 163-171.
- Shaw, W. C., Semb, G., Nelson, P., Brattström, V., Mølsted, K., & Prah-Andersen, B. (2000). *The Eurocleft Project 1996–2000. Standards of*

- Care for Cleft Lip and Palate in Europe*. European Commission Biochemical and Health Research. Amsterdam: IOS Press.
- Shprintzen, R. J., Siegel-Sadewitz, V. L., Amato, J., & Goldberg R. B. (1985). Anomalies associated with cleft lip, cleft palate, or both. *American Journal of Medical Genetics*, 20, 585-595.
- Sinko, K., Jagsch, R., Prechtel, V., Watzinger, F., Hollmann, K., & Baumann, A. (2005). Evaluation of esthetic, functional, and quality-of-life outcome in adult cleft lip and palate patients. *Cleft Palate-Craniofacial Journal*, 42, 355-361.
- Slifer, K. J., Amari, A., Diver, T., Hilley, L., Beck, M., Kane, A., et al. (2004). Social interaction patterns of children and adolescents with and without oral clefts during a videotaped analogue social encounter. *Cleft Palate-Craniofacial Journal*, 41, 175-184.
- Sommerlad, B. C., Mehendale, F. V., Birch, M. J., Sell, D., Hattee, C., & Harland, K. (2002). Palate re-repair revisited. *Cleft Palate-Craniofacial Journal*, 39, 295-307.
- Strauss, R. P., & Broder, H. (1991). Directions and issues in psychosocial research and methods as applied to cleft lip and palate and craniofacial anomalies. *Cleft Palate-Craniofacial Journal*, 28, 150-156.
- Strauss, R. P., Broder, H., & Helms, R. W. (1988). Perceptions of appearance and speech by adolescent patients with cleft lip and palate and by their parents. *Cleft Palate Journal*, 25, 335-342.
- Thomas, P. T., Turner, S. R., Rumsey, N., Dowell, T., & Sandy, J. R. (1997). Satisfaction with facial appearance among subjects affected by a cleft. *Cleft Palate-Craniofacial Journal*, 34, 226-231.
- Topolski, T. D., Edwards, T. C., & Patrick, D. L. (2005). Quality of life: how do adolescents with facial differences compare with other adolescents? *Cleft Palate-Craniofacial Journal*, 42, 25-32.
- Turner, S. R., Thomas, P. W., Dowell, T., Rumsey, N., & Sandy, J. R. (1997). Psychological outcomes amongst cleft patients and their families. *British Journal of Plastic Surgery*, 50, 1-9.
- Van Lierde, K. M., Monstrey, S., Bonte, K., Van Cauwenberge, P., & Vinck, B. (2004). The long-term speech outcome in Flemish young adults after two different types of palatoplasty. *International Journal of Pediatric Otorhinolaryngology*, 68, 865-875.
- Vanryckeghem, M. (1995). The communication attitude test: a concordancy investigation of stuttering and nonstuttering children and their parents. *Journal of Fluency Disorders*, 20, 191-203.
- Warschawsky, S., Kay, J. B., Buchman, S., Halberg, A., & Berger, M. (2002). Health-related quality of life in children with craniofacial anomalies. *Plastic and Reconstructive Surgery*, 110, 409-414.

- Whitehill, T. L. (2002). Assessing intelligibility in speakers with cleft palate: a critical review of the literature. *Cleft Palate-Craniofacial Journal*, 39, 50-58.
- World Health Organization (2001). *International Classification of Functioning, Disability, and Health*. Geneva, Switzerland: World Health Organization.
- World Health Organization (2002). *Towards a Common Language for Functioning, Disability and Health*. Geneva, Switzerland: World Health Organization.
- Yorkston, K. M., Beukelman, D. R., & Bell, K. R. (1987). *Clinical Management of Dysarthric Speakers*. London: Taylor & Francis.

Summary in Swedish

Svensk sammanfattning

Det övergripande syftet med avhandlingen var att undersöka och beskriva hur personer födda med läpp-käk-gomspalt (LKG) upplever sitt tal och sin kommunikativa situation samt huruvida deras bild av talet korrelerar med talbedömningar gjorda av LKG-specialiserade logopedier.

Både kvantitativa och kvalitativa metoder har använts. Femtiofyra tioåringar födda med LKG-spalt besvarade ett test av kommunikativ attityd hos barn (CAT-S) och resultaten jämfördes med föräldrarnas svar på frågor kring barnets kommunikativa situation samt med resultaten av logopeders talbedömningar. Barnens medelvärden på CAT-S jämfördes också med medelvärden hos 10-åringar utan spalt. Vidare tillfrågades 35 yngre vuxna personer födda med LKG-spalt om hur nöjda de var med sitt tal efter avslutad behandling vilket jämfördes med logopediska talbedömningar. Slutligen beskrev 13 yngre vuxna som var födda med LKG-spalt och som bedömts ha haft avvikande tal vid sitt senaste besök hos logoped i LKG-teamet sin uppväxt med spalt, sitt tal och sin kommunikation i kvalitativa semi-strukturerade intervjuer. Intervjuerna genomfördes och analyserades enligt riktlinjer inom den kvalitativa metoden "Grounded Theory".

Tioåringarnas svar på CAT-S korrelerade statistiskt signifikant med majoriteten av variablerna i logopedernas talbedömningar men sambanden förklarade endast en mindre del av variansen. Den kommunikativa attityden var signifikant mer negativ hos 10-åringarna med spalt jämfört med referensdata från 10-åringar utan spalt. Spridningen inom gruppen med spalt var dock stor. Vuxna personers egen utvärdering av talet korrelerade inte med talbedömningarna som gjorts av logopedier. Analysen av intervjuerna resulterade i två separata huvudområden med separata kärnkategorier. Kärnkategorin *Att göra spalten begriplig* (Making sense of the cleft) beskrev processen att utveckla självbilden i relation till spalten och omfattade kategorierna Att forma sin attityd till spalten (Shaping one's attitude to the cleft) och Att hantera att man är annorlunda (Dealing with being different). Kärnkategorin *Att ta sig an sin kommunikation* (Taking charge of communication) beskrev hur man hanterade talet och omfattade kategorierna Att göra sig en bild av talet (Forming an idea of one's speech), Att förstå sin kommunikation (Learning about one's communication) och Att ta ansvar för kommunikationen (Taking responsibility for communication).

Många barn och vuxna födda med LKG-spalt som konstaterades ha ett tal som avviker från normen vid den logopediska talbedömningen var själva nöjda med

sitt tal och beskrev en välfungerande kommunikation i de flesta situationer. Enbart logopediska talbedömningar säger således inte så mycket om personernas kommunikativa deltagande i samhället. Också personernas egen inställning till tal och kommunikation behöver därför undersökas mer strukturerat. Att tillföra information om personens egen bild av sin kommunikativa delaktighet skulle vidga det logopediska perspektivet från att bara bedöma talet till att också uppmärksamma hela den kommunikativa situationen på ett mer strukturerat sätt. Detta fyller ett behov både inom klinisk verksamhet och inom forskning kring personer födda med LKG-spalt.