Mental Health in Children Undergoing Reconstructive Surgery

Studies on Self-Esteem and Social Interaction

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Abstract

While the functional and anatomical aspects of reconstructive surgery in children with leg length inequality (LLI), prominent ears (PE) and cleft lip and palate (CLP) have been studied in detail, the psychological aspects of surgery have been less explored. The benefit of a changed appearance and function on self-esteem and ability to social interaction are other areas where information is lacking. The aim of this thesis is to examine, during the process of reconstructive surgery, the mental health, self-esteem and social interaction of children with defects in appearance and function.

Children, aged 6-16 years, with LLI (n=27) and PE (n=31) were invited to participate in interviews and psychological assessments by filling in a battery of questionnaires and tests (depression; anxiety; self-esteem; cognitive ability; and behaviour) before Ilizarov and otoplasty surgery and one year after. Parents filled in a child symptom check list and a state and trait anxiety questionnaire. Another six adolescents with CLP and their parents participated in interactive interviews with the aim of identifying relevant psychological issues for individuals with this condition. These issues were subsequently used to create new questionnaires. Being different, the development of self-esteem and social interaction were the central themes of the questionnaires designed after the interview study. The new questionnaires were explored in a retrospective study on other adolescents (n=26) with CL/P and their parents. Beck’s Youth Inventories (BYI) was used as comparative data.

The LLI group had significantly lower mental health and self-esteem scores than the control group before surgery. The leisure activity level in both patient groups was low according to parents’ report before surgery. The mental health scores of both patient groups (LLI and PE) were improved after reconstructive surgery, but self-esteem was not affected.

The questionnaires for CL/P patients proved to be useful in the exploration of self-esteem from a developmental perspective and in the search for strengthening factors of social interaction. Self-esteem was average or high on group level for adolescents with CL/P compared to BYI measure. Parents rated their adolescents to have higher self-esteem than the adolescents themselves. Females had a less positive development of self-esteem, there was an interaction effect between the female and male patients’ evaluation of self-esteem by higher age.

Even a minor appearance defect like PE may affect mental health negatively. There were no adverse psychological effects one year after surgery in LLI and PE patients, rather, there were signs of improved mental health. Adolescents with CL/P have an anticipated risk of more mental health problems and especially those individuals who have been bullied. Cleft teams and Child and Adolescent Psychiatric Clinics should be attentive and offer psychological support to those individuals most affected by their conditions.

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“Our shortcomings are our eyes with which we see the ideal”
Friedrich Nietzsche (1844-1900)
List of Papers

This thesis is based on the following papers, which will be referred to in the text by their Roman numerals:


IV. Niemelä J B, Skoog V, Henriksson T-G, Sundelin Wahlsten V. Self-esteem and social interaction in adolescents with CL/P. [Submitted for publication].
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Exploring Self-Esteem and Social Support in Adolescents with CLP in the Context of Secondary Correction of Lip/Nose (Paper III) ........28
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Abbreviations

ANOVA Analysis of Variance
BYI Beck’s Youth Inventories
CDI Children’s Depression Inventory
CLP Cleft Lip and Palate
CL/P Cleft Lip with /or without Cleft Palate
FI-CLP Family Interaction – Cleft Lip and Palate
ITIA I think I Am – “Jag tycker jag är” – Test of self-esteem
LLI Leg Length Inequality
LL Leg Lengthening
PE Prominent Ears
RCMAS Revised Children’s Manifest Anxiety Scale
SPIQ Speedy Performance Test on Intelligence
YQ-CLP Youth Questionnaire – Cleft Lip and Palate
Introduction

The main focus of the thesis was to investigate the psychological consequences of defects in appearance and function in childhood. A further aim was to examine how reconstructive surgery affected mental health, self-esteem and social interaction. While the functional aspects of the reconstructive surgery have been studied in detail, the psychological aspects of the surgery have been less explored (Eiserman, 2001). The impact on mental health has been questioned when there are demanding surgical procedures and lengthy treatments.

The benefit of a changed appearance and function on self-perception, self-esteem and ability to social interaction are other areas which have been studied, but with diverse outcomes. In modern society more and more attention is paid to appearance and to an adjustable social behaviour. This can create demands from parents and adolescents on the health care system for more refined reconstructive surgery. More knowledge is therefore needed to meet these demands and offer children what is best for them. Feeling different is a psychological dimension in childhood and adolescence that is even more intense if there is a deformity in both function and appearance. In studies of self-esteem in adolescents with CLP, de Andrade et al. (2001) found that these young people performed differently from those without cleft, that is, they presented lower self-esteem scores. Cleft lip can also be a risk factor for mental health problems (Marcusson, 2002) and for suicide in adults according to Herskind et al. (1993).

In the clinical practice of the Pediatric Orthopaedic Department and the Child and Adolescent Psychiatric Clinic, a collaborative project initiated the project to examine the mental health of leg lengthening patients. In order to compare if children’s mental health was affected also by a minor deviance in appearance and a less demanding surgical procedure, otoplasty patients were included in the studies as a comparison group. Data from this study raised some interesting questions about condition specific self-esteem in children with facial deformities. In search of a new understanding of how the condition of Cleft Lip and Palate (CLP) affected adolescents, this patient group was also invited to participate in the study of self-esteem and social interaction.

Thus, three patient groups with a variety of deformity in appearance and function have been studied – Leg Length Inequality (LLI), Prominent Ears (PE) and Cleft Lip with or without Palate (CL/P) – in order to catch the dilemma of being different and the development of self-esteem in the context...
of reconstructive surgery. The diverse appearance problems are also interesting since the fact of a defect in the face can create more problems than a major orthopaedic problem with function and appearance might present (Harper, 1995).

The Effect of Reconstructive Surgery

Studies of orthopaedic and other surgical treatments demonstrate that psychological problems are associated with lengthy procedures, lack of information and support to parents, lack of counselling to patients and their parents, maladaptive coping behaviour, and child surgery at an inappropriate developmental level (Braun et al., 1995, Maffulli and Fixsen, 1996, Hesham et al., 1996, LaMontagne et al., 1997, Hägglöf, 1999). However, cognitive ability is considered a protective factor in studies of children at risk (Garmezy et al., 1984, Burnstein, 2003, Caffo and Belaise, 2003, Dyregrov, 2006). Lower levels of reported parenting stress were also related to better social skills in the child (Krueckeberg and Kapp-Simon, 1993). Children’s reactions to reconstructive surgery in general have not been studied to any great extent.

Leg Length Inequality Patients (LLI)

Severe maladaptive psychological reactions associated with the lengthy and demanding procedure of leg lengthening (LL) have been reported (Hrutcay and Eilert, 1990, Tjernström et al., 1990, Birch and Samchucov, 2004). Hrutcay and Eilert (1990) reported that a child’s reactions to leg lengthening could be anxiety, regression, dependant, guilt, and acting-out reactions, depression, anorexia and self-destructive behaviour during hospital care. It was understood as Adjustment Disorders or, according to DSM III, as Maladaptive Reactions to an Identifiable Psychosocial Stressor. No psychological assessment was conducted before surgery so we do not know whether there were preexisting difficulties behind the children’s reactions to LL.

The etiology of leg length inequality may also affect the reaction to surgery, as Tjernström explains:

The history of the leg length inequality may also affect the outcome, for instance if it has a congenital or traumatic etiology. Patients with a congenital shortening become accustomed to the slowly increasing inequality from childhood and have no experience of a life without this handicap, while a traumatic shortening is sustained suddenly and, despite optimal treatment, is difficult to restitute to a pre-fracture status (Tjernström, 1994).

Prominent Ears Patients (PE)

Children with prominent ears (PE) appears to be affected in their psychosocial adjustment but they do have problems with self-esteem when compared
with normal children and children with facial “Port wine stains” (Sherin et al., 1995). Despite the possibility to use psychological treatment, this is rarely provided for various reasons such as lack of practitioners and underdeveloped research base. Therefore in many places surgical treatments are the most commonly offered intervention. Opinions when this option should be present differ. For instance, there are surgeons who view surgical corrections as the patient’s right, even when there are minor deformities. One argument is that every child deserves good health and a sense of well-being (de Chalan, 1997).

Bradbury et al. (1992) found improved well-being in 90% of the children one year after ear correction. A small group (10%) of the children remained dissatisfied after surgery. They were children who were socially isolated prior to surgery. The authors suggest careful screening and referral back to the family doctor for the more distressed children. The majority of studies have found that reconstructive surgery on PE significantly reduces peer ridicule and increases self-esteem. (Bradbury et al., 1992, Horlock et al., 2005)

Cleft Lip and Palate Patients (CLP)
The psychological aspects in relation to CLP have been examined for decades with an assortment of aims and outcomes. On the one hand the focus has been on children’s own experiences of their appearance and what they think other people’s reactions are as well as a focus on social adjustment and social interaction. The effect of reconstructive surgery has usually not been in focus in CLP affected individuals, since the surgical interventions have been a medical necessity.

Satisfaction with appearance from both the child’s and parent’s perspective has been explored. Appearance might influence social interaction when the ability to show emotional expressions is diminished or deviant. Slifer et al. (2003, 2004) examined the ability of children to decode feelings in other people and to show feelings by facial expressions. Oral cleft children had a different way of expressing feelings compared to controls and there was an association with perceived social acceptance. Parent perception of their child’s social competence and the child’s report on self-perception of social acceptance was positively correlated for both groups. Many studies underline that children can adjust well psychologically, but their ability of good social adjustment is more varied (Campis et al., 1995, Turner et al., 1998, Broder et al., 2001).

Adult female patients have reported dissatisfaction with their aesthetic outcome (Sinko et al., 2005). Aesthetic gain can heighten self-perception and therefore the patient’s satisfaction with her facial appearance should move more into focus of the therapy of clefts, according to Landsberger et al. (2006). The task for the plastic surgeon is to weigh the patient’s psychosocial problems against the limitations and risks of surgery (Harris, 1982).
Mental Health and Self-Esteem

According to empirical research there is a positive correlation between mental health and self-esteem. A person with good mental health seems to have a positive attitude about themselves, but a person with bad mental health has a negative attitude. Self-esteem is a sensitive aspect of a person’s identity and personality (Ouvinen Birgerstam, 1985, Shek, 1998).

An association between mental health and self-esteem is documented in psychiatric clinical research. Depressed adolescents, e.g., seem to have low self-esteem (Kazdin, 1990). A stressful life condition does not automatically imply low self-esteem. Contrarily, some groups of otoplasty patients report high or very high self-esteem although they do have difficulties in handling frequent hospital visits and treatments, absence from school and loss of interaction with peers (Sherin et al., 1995). One reason could be the benefit of getting more time together with the parent when the child has to go to hospital, and also the parents’ efforts to care for the child. Meeting with hospital staff and other patients, with the same condition, might also encourage feelings of coherence and general sense of well-being.

In a study by Hunt et al. (2006) patients with CLP had higher depression scores and behaviour problems compared with controls, but there was no difference in terms of self-esteem. However, Cheung et al. (2007) found that adolescents with CLP had good relations with parents and no social anxiety but lower general and social self-esteem compared with controls. It is recognised that life conditions and development in themselves influence the psychological status of the child and possibly also their self-esteem (Cheung et al., 2007).

Defect in Appearance – A Stigma?

Because of the centrality of the face in human interactions and development, facial differences may be particularly stigmatizing (Cole, 1998). Studies regarding non disabled young people’s attitudes toward their peers with disabilities show that non disabled young people prefer those with mobility limitation to those with facial differences (Harper, 1986, Harper, 1995). Adolescents with facial differences confront significant challenges to their own self-identity, while experiencing a higher quality of life from relationships, possibly from their need to negotiate and maintain close family support.

Characteristics other than the direct visual impact of a defect in appearance may also influence the degree of stigmatization experienced. For instance, the ability with which a CLP affected individual is perceived to be handling their imperfect appearance can favourably alter an outsider’s judgement of them (Stricker et al., 1979). There is some speculation that milder disfigurements cause as much anxiety as, if not more than, severe disfigurements do (Sherin et al., 1995, Moss, 2005, Landsdown et al., 1991). The social response towards individuals with milder defects is less predict-
able than that towards severely disfigured individuals. It is the unpredictability of the social response that is thought to raise anxiety levels. CLP affected individuals who are dissatisfied with or who are unrealistic about their appearance need to be identified. Once found, these individuals “at risk” of developing low self-esteem, could be offered some social skill training in order to improve their self-confidence and increase their self-esteem (Rumsey et al., 1986, Kapp-Simon, 1995, Turner et al., 1998, Kapp-Simon, 2004, Kapp-Simon et al., 2005, Lowegrove and Rumsey, 2005).

The Dilemma of Being different

One obvious reason for reconstructive surgery is a child’s problem with motor or speech function, but also a deformity in appearance can motivate an intervention. Being different might also interfere with capacity for social interaction. CLP affected individuals have reported inhibited behavior (Broder, 2001, Broder et al., 2001) as well.

When there is a minor deviance, such as ear deformity, there can be difficulties in social interaction. In studies 12 months and 2.2 years after reconstructive surgery of prominent ears, an enhanced social life and leisure activity were demonstrated (Bradbury et al., 1992, Horlock et al., 2005). This change in activity was associated with a better self-confidence and less psychosocial problems.

Reconstructive Surgery and Self-Esteem

There is still much to explore regarding the concept of self-esteem, especially in individuals affected by deformities. Earlier studies usually report low self-esteem and the need for surgery in order to improve self-perception. Adolescents with CLP are one group of patients reported to have lower self-esteem than other adolescents (de Andrade and Angerami, 2001).

The development of reconstructive surgery has been successful and nowadays no physical dysfunction appears among adults, but there are still some residual stigmata to explore. For instance, smaller defects in appearance or speech can make life difficult in social situations when meeting unknown peers. Studies have demonstrated that surgical intervention in CLP patients has often resulted in higher levels of self-confidence, self-esteem, and a better self-concept (Lefebvre and Munro, 1978, Clifford, 1987)

Definitions of Self-Esteem

Self-esteem can be described within the frame of self-concept, which is a broader definition. From the beginning self-esteem was referred to as a one-dimensional concept. In accordance herewith, self-esteem was operational-
ized and measured as a kind of global self-evaluation (Marsh and Hattie, 1996). After a study by Shavelson et al. (1976) researchers started to examine self-esteem as a complex and multidimensional model of the self-concept. They explained the model with global self-esteem on top and with layers underneath with, e.g., an academic, social, emotional, and physical self-concept – and further on layers including appearance and physical ability (Lindwall, 2004).

In the research area of CLP the definition of self-esteem by Cheung et al. (2007) is: The individual’s experience of his/her own value and competence. Furthermore, that self-esteem is related to many aspects of oneself and one’s relation to others (Hunt et al., 2006, Cheung et al., 2007, Topolski et al., 2005). The importance of the relationship with parents for children to have a sense of a self and psychological well-being is documented by Stern (1985) and Shore (2003). Another way to describe it is that the self-esteem can be buffered by parents. Cheung et al. (2007) found that self-perception of general and social self-esteem in adolescents with CLP was low, while the parents rated them to have higher self-esteem than a non-affected group of children. Parents usually perceive and report externalized behaviour while adolescents report internalized problems.

Except for the difference in children’s and parents’ ratings there is also a difference between patient groups. Patients with chronic diseases were found to cope better than adult patients with CLP. This could be attributed to the fact that cleft deformity is considered to be an anomaly rather than a symptom (Cheng et al., 2000).

Simis et al (2001) refer to the fact that psychological measures usually demonstrate only mild or no psychological problems. By contrast, clinical reports, semi-structured interviews and study specific questionnaires reveal that child and adolescent patients encounter a range of appearance related problems, such as being teased, feeling inferior and rejected or self-conscious, being upset about their disfigurement, and feeling depressed. The use of instruments designed to assess specific areas of self-concept rather than more global measures are also justified (Gussy and Kilpatrick, 2006). Broder (2001) also emphasizes the need to develop specific condition formulas, including self-esteem measures for young people with CLP, which might enhance the possibility to screen for psychological distress. Having a cleft of the lip and palate has a specific rather than a broad association to psychosocial adjustment.

In a “State of the Art” article Endriga and Kapp-Simon describe the complexity of the concept of self-esteem:

Researchers have frequently examined self-concept as an important variable in the adjustment of children with craniofacial anomalies. Depending on the instrument used for assessment, self-concept includes a number of different constructs that are considered to influence the way an individual feels about
himself and fashions relationships with others. Frequently occurring constructs include self-assessment of appearance, behavior, social acceptance, cognitive functioning, and self-worth (Endriga and Kapp-Simon, 1999).

Ouvinen-Birgerstam (1985) has developed the instrument called “I think I am” (ITIA) in order to measure self-esteem in children, 7-15 years of age. ITIA covered most of the factors of self-esteem that the author of this thesis intended to measure: how the individual values her/his body, abilities, mental health, relations to parents and others.

I Think I Am (ITIA); intends to examine children’s self-evaluation. The ITIA has 72 items, in 5 separate scales, and is standardized on Swedish schoolchildren. The ITIA is inspired by several well documented self-esteem measures, for instance Self-Esteem Inventories (SEI); Piers-Harris’ Children’s Self-Concept Scale (PHSCS) and Rosenberg’s self-esteem questionnaires (RCE) Coopersmith, 1989, Piers, 1996, Rosenberg, 1965).

The ITIA is being widely used in Sweden in order to study self-esteem in different diagnose groups of children with chronic conditions and healthy children (Engström, 1991, von Essen et al., 2000, Räty et al., 2005, Flodmark, 2005, Kalifa, 2006, Lindfred et al., 2008, Daud et al., 2008).

The Process of the Thesis

This thesis consists of four studies (Paper I-IV). In the first two studies (Paper I and II) the ITIA was used to examine the self-esteem of patients who had undergone reconstructive surgery (Niemelä et al., 2007, Niemelä et al., 2008). Since no significant effects on self-esteem were reported, the need for new understanding and, eventually, new measures was evident.

An interactive interview (Paper III) was conducted with a sample of adolescents with CLP about their self-esteem and its development over childhood and adolescent years. Retrospective questions on self-esteem and ideas of how social support affected self-esteem and ability for social interaction were explored. Important themes were also how their self-esteem influences the adolescent’s decision-making regarding additional surgical interventions.

After the interview study of the 6 adolescents with CLP and their accompanying parents, material for new questionnaires was collected. After careful consideration of the current research area on CLP and self-esteem measures and the authors’ clinical experiences, the new questionnaires Youth Questionnaire-CL/P (YQ-CL/P) and Family Interaction-CL/P (FI-CL/P) were developed. The intention was to find those items concerning self-esteem and social interaction of interest for CLP affected individuals.

The themes that were to be examined (Paper IV) were the following. First there is the developmental process when the child becomes aware of the defect in appearance and function and eventually starts to reflect upon the condition. Measuring self-esteem at one occasion did not seem to catch the
impact of reconstructive surgery. Instead retrospective questions of how the
adolescents’ self-esteem had changed during the process of surgical proce-
dures were to be explored. The targeted ages of 6, 8-10 and 13-19 years to
ask people about their self-esteem were also the ages when important surgi-
cal interventions had been performed in CLP patients.

Second there is the need to examine the themes emerged from the inter-
view study (Paper III) and to learn from adolescents’ answers to the ques-
tions that were designed after that study. For instance their experience of
CLP, what had strengthened self-esteem, what support they appreciated from
people around, their social interactions, the defect in appearance and motiva-
tion for surgery. To search for reason and motivations for self-esteem is re-
lated to the self-esteem models that have been examined by Byrne and
Aims of the Thesis

- To investigate the mental health of children undergoing leg lengthening.
- To ascertain possible differences in the effect of reconstructive surgery on mental health and self-esteem when the child has a minor defect in appearance and function – as, e.g., prominent ears in comparison to leg length inequality.
- To explore self-esteem and social support in adolescents with CLP in the context of secondary correction.
- To explore self-esteem and social interaction in adolescents with CL/P by means of a new condition specific questionnaire.
Methods

Participants

*Paper I*

**Leg lengthening patients (LLI).** During 1997-2006, 42 children, aged 6-16 years underwent LL. The study included 27 patients (13 girls and 14 boys, average 11.3 years), with leg length inequalities (LLI) and limb deformities. The patients underwent leg lengthening (LL) using the Ilizarov technique, at the Pediatric Orthopedic Department, Uppsala University Hospital.

Dropouts: 8 were excluded because of preoperative problems not related to LLI; 4 patients were excluded because they missed the preoperative psychological tests; and 3 patients were excluded because they underwent their second or third LL.

LLI, whether congenital or acquired, is common (Menelaus, 1991). At Uppsala University Hospital, patients have been treated for LLI since 1984 with different reconstructive surgical procedures. LL is recommended if the expected LLI will be more than 6-7 cm when the patient is fully grown. LL can be performed from the age of six according to Rehnberg (2007). The treatment is lengthy and demanding with frequent complications and postoperative pain. The convalescence is usually long.

*Control group.* Twenty-seven school children, matched for age and sex with the LLI group, were recruited from one school area in Uppsala.

*Paper II*

**Prominent ears patients (PE).** A total of 54 children, 6-16 years of age, underwent otoplasty at the Plastic Surgery Clinic, during 2000-2006. Forty-two children participated in the study.

Dropouts: 12 patients that had received information about the study did not take part for the following reasons: six declined; two could not wait for their turn to be interviewed; and four could not be examined because the psychologist was not on duty.

Indication to operate was a distance greater than 16 mm between helix and cranium, i.e., a clear medical deviation.

*Comparison group.* Twenty-one LLI patients, matched for age and sex.
Otoplasty is one of the most frequently performed aesthetic, surgical procedures in children and adolescents (Burnstein, 2003). Prominent ears are relatively common with an incidence of 5% in the Caucasian population (Adamson and Strecker, 1995). The condition is inherited as an autosomal dominant trait and is commonly caused by a combination of two defects: underdevelopment of antihelix folding and overdevelopment of the conchal wall (Bhatti and Adeniran, 2006).

Open Otoplasty (Nordzell, 2000) is performed at the Daycare Unit, Plastic Surgery Clinic, Uppsala University Hospital. The surgical procedure is conducted under local anaesthesia and takes 1-2 hours. The child can leave hospital the same day and return to school in a few days.

**Paper III and Paper IV**

Cleft lip/palate patients (CL/P). Adolescents of 13-19 years of age that underwent secondary correction of lip and nose were consecutively invited to take part in the studies.

Six CLP patients (3 males and 3 females) took part in the interview study (III) in 2005 and 26 adolescents with CL/P (17 males and 9 females) participated in the main study (IV), at the Plastic Surgery Clinic, Uppsala University Hospital, during 2005-2008.

Dropouts from study IV: 3 males declined to participate; 3 males and one female did not want surgery; two (one male and one female) could not take part because of logistic reasons or psychologist not on duty.

There was no difference between participants and non-participants on any demographic variable.

The incidence rate in Sweden is about 1.8/1000 newborn children (Henriksson, 1971, Robert et al., 1996). Most children with any kind of CLP, having undergone an operation for Cleft lip, will need secondary surgery later on in order to approve the appearance of lip and nose. These interventions are usually not performed before the patients have reached their teens but they can also be conducted on adult patients. This study focuses on the adolescent patient’s expectations, when it is time for plastic surgery to improve their appearance: the secondary correction of lip and nose.

**Procedures**

The studies were performed at Uppsala University Hospital, in the Paediatric Orthopaedic Department, the Plastic Surgery Clinic, and the Child and Adolescent Psychiatric Clinic.
In *Paper I* medical data were examined by the surgeon (BT) via the clinical records on diagnosis, the surgical procedures, complication rate and type, days of hospitalization, and gained length.

Psychological data were apprehended by the psychologist (BJN) from interviews, test, and self-rating scales while psychological assessments with child and parents were conducted at the Child and Adolescent Psychiatric Clinic. The preoperative assessment took place on average 19 days before and the follow-up assessment 12-13 months after leg lengthening. The remodelling of the new bone to an appearance that resembles the surrounding bone takes at least one year (Tjernström et al., 1992).

The control group data were obtained from one assessment using the same procedures as for the patients.

The psychological assessment for *Paper II* underwent the same procedure as did the one for the study in *Paper I*. The only difference was that the pre-surgery assessment with the PE patients was conducted at the Day-Care Unit, the Plastic Surgery Clinic one week before the otoplasty.

Comparison between the clinical groups (LLI and PE) and the matched control group was made at a baseline, but only the clinical groups were included in the post-treatment phase.

Medical data for the PE patients were obtained and examined by the surgeon (AH).

In *Paper III* and *Paper IV* the psychological measurement data were obtained by the psychologist (BJN) via the interviews with and self-reports of adolescents with CLP and their parents at the in-patient ward, the Plastic Surgery Clinic, the day before secondary correction of lip/nose.

The adolescent was asked whether they wanted their parents present during the interview (*Paper III*) to create as safe a situation as possible for the patient in a strange milieu where the surgical intervention might provoke anxiety. In the study III, the surgeon also took part in the interactive interview, the surgeon was included to provide a setting where it was possible to talk about genetic concerns such as having a baby of your own with CLP and other medical issues.

Medical data for the CLP patient in the preliminary study were examined by the same surgeon (T-G H).

All interview material in *Paper III* was analyzed separately, then discussed by the authors, and finally collected into crucial themes and concepts. This qualitative method is described by Shotter (1996). The validity of the material was interpreted by the authors’ discussion and built on professional experiences according to the ideas of assessing quality in qualitative research (Mays and Pope, 2000).

In *Paper IV* the adolescent was invited with the parents to the interview together and then separated to fill in the forms with the psychologist (who is not in the cleft team) who is present to help and explain the forms.

The surgeon (VS) examined the medical data regarding patients in paper IV.
Internal academic review and external ethics approval were obtained for the studies.

Measures

Paper I and Paper II

The following psychological measurements and semi-structured interviews have been used in Paper I and Paper II in order to assess aspects of the mental health in the LLI and PE patients:

**The Children’s Depression Inventory (CDI).** The CDI is a 27-item, self-reported, symptom oriented scale that was designed for school-age children and adolescents. Each item consists of three choices, keyed from 0-2 in the direction of increasing severity. Thus, the total score ranges from 0-54. A score of 9 points or less is considered normal range (Kovacs, 1982).

**“I Think I Am “ (ITIA).** A self-rating scale of self-esteem was measured by two versions of ITIA. ITIA for children aged 7-9 consists of 32 items with a total value range of -32- +32. Norms for total points have shown from 15.4 (SD 10.24) – 22.37 (SD 7.73). Children under 10 years of age were also asked to draw a picture of themselves and the people who loved them best.

ITIA for children 10 years or older includes 72 items and can reach a value from -144–+144. In norm groups M values in between 60.50 (SD 33.4) – 74.90 (SD 30.0) are presented, and in chronically diseased groups 47.40 – 62.40. Raw scores are converted to stanine scale 1-9 (Ouvinen Birgerstam, 1985).

**Revised Children’s Manifest Anxiety Scale (RCMAS).** A scale measuring a child’s anxiety within the last two weeks. RCMAS includes 28 statements which the child answers yes or no to. The total value can vary between 0 and 28. In norm groups M values in between 18.1 (SD 6.3) – 11.9 (SD 6.4) are reported. (Reinolds and Richmond, 1978).

**“Speedy Performance Test on Intelligence” Snabb performancetest på intelligens (IQ) (SPIQ I och II).** This test can be administered from 2 years to adult age. Norm groups: 2-16 years. The task is to associate words with pictures. The test examines reasoning and inductive thinking. There are two equivalent parts of the test (I and II); the first one was administered before and the second one a
year after surgery. The raw scores 0-60 are converted to a stanine score 1-9. (Rydberg and Höghielm, 1985).

**State-Trait Anxiety Inventory, form X (STAI).** Measured parents’ anxiety when a child was going to have reconstructive surgery and again one year after surgery (Spielberger, 1983).

The State-Trait Anxiety Inventory has been used extensively in research and clinical practice since it was introduced in 1966. The scale has been used where there has been a need to distinguish between anxiety as a transitory emotional state and individual differences in anxiety proneness as a relatively stable personality trait. The sensitivity of the S-Anxiety scale to environmental stress has been repeatedly demonstrated in research on emotional reaction to surgery.

**Self-Perception Leg Length Inequality/Prominent Ears.** A semi-structured interview formula developed by the first author (unpublished manual by Niemelä J) in order to gain more direct and qualitative information from the child concerning his/her experiences of LLI and PE. The formula was based on clinical experiences and systemic theory inspired by Andersen (2003).

Post-surgery questions concerning overall outcome and satisfaction, which were rated on a 1-5 point scale (extremely satisfied, moderately satisfied, no opinion, moderately dissatisfied, extremely dissatisfied).

Another follow-up question, which concerned pain and other complications, was: “Do you think it was worthwhile?”

**Paper III**

The following psychological measure was used in Paper III, the preliminary with the CLP patients undergoing secondary correction of lip/nose:

**A Semi-Structured Interview.** The interview was constructed for adolescents with CLP by the first author (Niemelä J, 2004), inspired by post-modern thinking on qualitative interviews (Gergen 1994, Katz and Shotter, 1996, Kvale, 1997, Polkingthorne, 1983, Andersen, 2003).

The questions were related to experiences of living with CLP and how CLP affected feelings, thoughts and behaviour. What expectations did adolescents have on reconstructive surgery (secondary correction of lip/nose)? How did they cope with difficulties in life and were there differences according to gender? What had been the best support? How was their self-esteem in different stages of life?

The adolescents were told that it was an interactive interview and their ideas and experiences were of utmost importance for further
studies. Parents were also invited to take part, if the adolescents so decided.

All interviews were video/taped and transcribed.

**Paper IV**

The following psychological measures and semi-structured interviews were used in Paper IV in order to assess aspects of the mental health in the CL/P patients.

Since the CL/P patients were older than the LLI and PE patients, another psychological measure of mental health, the BYI, was used.

**Beck’s Youth Inventories (BYI). Beck ungdomsskalor.** An instrument designed to measure different aspects of mental health in young people aged 9-18, by self-assessment of anxiety, depression, anger, antisocial symptoms, and self-esteem in different subscales (Tideman, 2004).

**Youth Questionnaire-CLP (YQ-CLP)** (unpublished manual by Johansson Niemelä). YQ-CLP contained 24 items in different formats, spanning from 2-9 categories on social interaction, patients’ experiences and expectation on reconstructive surgery and second correction of lip/nose, changing the perspective by thinking how it would be to have a child of your own with CLP.

The patients were also requested to assess their self-esteem on an analogous scale. The psychologist explained the procedure to the patients. ”Estimate your self-esteem on a scale between 1 and 9; 1 if you dislike everything about yourself and 9 if you find everything about yourself really good”. “What was your self-esteem at different ages, say 6, 8-10 and 13-19?”

**Family Interaction-CLP (FI- CLP)** (unpublished manual by Niemelä J). The parent questionnaire had two versions: one for the mother and one for the father. The questionnaire had 29 items in different formats, spanning from 2-9 categories.

The items of the two questionnaires, YQ-CLP and FI-CLP, are related to six dimensions: personal data, social interaction, affects, surgical interventions, self-esteem, and support intervention.

The items had firm response alternatives, but space was also provided for comments and concerns.

The following themes of interest were examined concerning self-reflection and social adjustment: self-esteem and male and female per-
spective on CLP. The importance of parent support, caring, and social interaction was also explored.

Both questionnaires were specially designed for this study by the first author (unpublished manual by Niemelä J, 2005).

The questionnaires were based on two studies (Papers I and II) on children’s mental health and self-esteem when undergoing reconstructive surgery and the interview study (Paper III) (Niemelä et al, 2007, 2008).

The questionnaire forms are outlined in Appendix I and II.

Statistical Methods

In Paper I mean differences were analyzed with paired t test. A significance level of 5% was used; however, because of multiple comparisons, exact p values and degrees of freedom (df) were also presented. The 5% level was used to balance the low power of the study with the risk of missing potential differences. However, the readers are encouraged to view significance levels above the 1% level with caution. All analyses were performed using SPSS 11.0 software.

In Paper II non-parametric tests were used because of relatively small sample sizes and the fact that measures of skewness and kurtosis indicated violation from normality for some of the variables. Group differences were analyzed with the Wilcoxon signed rank test and the Friedman test. Because parametric tests yielded corresponding outcome, means and standard deviations were also presented. A significance level of 5% was endorsed. All analyses were performed using SPSS 11.0 software.

In Paper IV differences in proportions were analyzed with Chi-2 test. Spearman’s correlations coefficient was used for testing the strength of association between variables. Pairwise differences were examined with Wilcoxon signed ranks test and Friedman test. Repeated measure ANOVA was used for measuring interaction effects over time. A significance level of 5% was used. All analyses were performed using SPSS 13.0.
Results

Mental Health and Reconstructive Surgery (*Paper I*)
The aim of *Paper I* was to examine the mental health of the children who underwent reconstructive surgery with the Ilizarov Leg Lengthening (LL) technique. The study was performed on a sample of 27 Leg Length Inequality (LLI) patients aged 6-16. The patients were examined before surgery and one year after. The mental health of the patient group compared with that of a matched group of healthy school children demonstrated lower mental health and self-esteem according to self-reports and parent reports before surgery. Post-surgery psychological measures of depressive and anxiety symptoms were significantly reduced, as well as the parents’ reports on the CBCL subscales: Aggressive behaviour, Attention and Externalization problems. Parents’ state anxiety (STAI) was also reduced.

The effect of gender was not significant in this study. However, the anxiety and depression scores were higher in the age group of 10-12. Parents also reported more problems on the CBCL in this age group.

Patients, and especially parents, reported pain, psychological discomforts, complications, and restrained function during LL. Satisfaction with surgery was high, although the patients reported that the surgery could be too trying for young children. There were, however, no adverse psychological effects one year after the reconstructive surgery with the Ilizarov technique but, rather, there were signs of improved mental health.

The Effect of Reconstructive Surgery on Mental Health when the Child has a Minor Defect in Appearance and Function Compared to a Major Defect (*Paper II*)
A sample of 31 Prominent Ears (PE) patients aged 6-16 was examined before surgery and one year after. Twenty-one youngsters could be matched with the leg lengthening group and the control group. The motivation of the PE children to be operated on was pain, teasing and feelings of being different. The patients’ rating of satisfaction with the result of the otoplasty surgery was high. The psychological measures of the mental health in the 21 PE patients before surgery were close to the results of the group of healthy
school children. The LL group had significantly higher depression and anxiety scores. Self-esteem was also the lowest in the LL group. After surgery the parents of the PE group reported improved behaviour on CBCL total problem score on the scales of Somatic complaints and Attention problems. Interestingly, parents reported less activity at leisure time in both patient groups than in the control group.

The effect of gender and age was not significant in either study I or II. However, the self-esteem was not affected in the patient groups according to the ITIA. Nor was this expected, since in the interviews the patients often expressed a change in their life situation after surgery and they felt safer and valued themselves higher. This motivated for further research on self-esteem and for finding condition specific measures for patient groups with defects in appearance and function.

Exploring Self-Esteem and Social Support in Adolescents with CLP in the Context of Secondary Correction of Lip/Nose (Paper III)

In study III 6 adolescents with CLP were invited to an interactive interview with their plastic surgeon and a psychologist. The aim of the interview was to explore the experiences of living with CLP: the effect on self-esteem and social interaction. Parental support were also focused on. The interviews were conducted in an in-patient setting, the day before secondary correction of lip/nose. Secondary correction of lip/nose is a surgical intervention that intends to adjust appearance toward a normal standard. The request for surgery differs among patients and the reason for this is not clear. One reason could be self-esteem and social support. The fear of surgery, having to be absent from school, etc. could be deciding factors for not wanting to be operated on as the decision-making is up to the adolescent him-/herself.

The main outcome was the themes: process of change, dependence of parental support, perspective taking, and self-esteem described in a more affective way than expected.

The outcome of the interviews with adolescents and their parents became the base for two new formulas, Youth Questionnaire-CLP (YQ-CLP) and Family Interaction-CLP (FI-CLP), which were intended for the main study (Paper IV) of self-esteem and social interaction in adolescents with CL/P.
Exploring Self-Esteem and Social Interaction in Adolescents with CL/P with the New Condition Specific Psychological Measures (Paper IV)

A sample of 26 CL/P adolescents, 13-19 years of age, took part in an explorative study at the Plastic Surgery Clinic, Uppsala University Hospital, in order to analyze self-esteem and social interaction by means of two new condition specific formulas YQ-CLP and FI-CLP.

Parental support was chosen by 80% of the patients as the most important support for coping with CLP.

The self-esteem was above median level for this group of adolescents; see Table 1. Parents rated their children to have higher self-esteem when they grew older; see Table 2. However, females reported a diminished level of self-esteem from primary school to secondary school; see Figure 1. When comparing the results of BYI’s self-esteem item we found a satisfying correlation of 0.57 (p<0.001) with the YQ-CLP. Using the new formulas demonstrated a new understanding of the self-esteem under development, also to differentiate between parent-adolescent estimation of self-esteem.

Parents reported that crises were common in their spouse (husbands 47% and wives 25%) when the child was born with CLP but usually they were left without support. Parents also reported good attachment to their child, and extra bonding because of extended time alone in hospital.

The overall measure of BYI for the CLP group demonstrated a profile more like a standard group than clinical groups with psychiatric diagnoses; see Figure 2. However, according to BYI, mental health was affected by CLP, and especially so by the ones who had been bullied. After crosstabulation of YQ-CLP and BYI data there was a trend between those adolescents that wanted to change their appearance as first choice, versus those with other first choices, and BYI scales on anxiety ($\chi^2$ 5.2, p= 0.08) and depressions ($\chi^2$ 5.3, p= 0.07).

Future Intervention suggested by the CL/P patients

Half of the CL/P adolescents’ choices were group-setting interventions: either therapeutic talks or the training of new social abilities as future interventions at the Plastic Surgery Clinic.

Supporting the parents was suggested by the adolescents, but not family consultation.

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Tables: Self-Esteem Estimations by Adolescents with CL/P and their Parents

**Table 1.** Patients with CL/P answers to the question “How was your self-esteem when you were 6 years, 8-10 years and 13-19 years of age?”; the result was on a scale 1-9:

<table>
<thead>
<tr>
<th>Age</th>
<th>M</th>
<th>(SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>6 years of age</td>
<td>6.8</td>
<td>(2.0)</td>
</tr>
<tr>
<td>8-10 years of age</td>
<td>6.5</td>
<td>(1.7)</td>
</tr>
<tr>
<td>13-19 years of age</td>
<td>6.6</td>
<td>(1.9)</td>
</tr>
</tbody>
</table>

**Table 2.** Parents’ reports on the question “Rate the self-esteem of your child when it was low and how the self-esteem is now.”

<table>
<thead>
<tr>
<th>Parents’ estimation of children’s self-esteem</th>
<th>Self-esteem when it was lowest</th>
<th>Self-esteem at present time</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parents n = 25</td>
<td>M: 5.3 (SD 1.4)</td>
<td>M: 6.8 (SD 1.4)</td>
</tr>
<tr>
<td>Fathers n = 11</td>
<td>M: 4.8 (SD 1.2)</td>
<td>M: 7.0 (SD 1.1)</td>
</tr>
<tr>
<td>Mothers n = 19</td>
<td>M: 5.3 (SD 1.5)</td>
<td>M: 6.8 (SD 1.5)</td>
</tr>
</tbody>
</table>
Figure 1. Gender and self-esteem.

Interaction effect between 8-10 and 13-19 years of age
\( F=4.4; p<0.05 \)

Figure 2. Beck Youth Inventories - Mean values in three groups: Standardized sample, 149 psychiatric patients and 25 CL/P patients.
General Discussion

Mental Health in Children Undergoing Reconstructive Surgery

From Papers I, II and IV we found that the patient groups have a mental health profile close to the normal population. This was informative since severe psychiatric reactions have been reported in connection with other leg lengthening studies (Birch and Samchucov, 2004, Hrutcay and Eilert, 1990, Tjernström et al., 1990).

The LLI and PE patient groups were compared to a control group of healthy school children, matched on age and gender, for pre-surgery baseline measures. The LLI group had more problems according to both self-reports (CDI; RCMAS; ITIA; YSR) and parent reports (CBCL) compared to the control group.

The mental health for both patient groups did not deteriorate by the intervention of reconstructive surgery but, rather, their mental health was improved. Parenting stress was high in the LLI patient group, but was reduced one year after surgery. The parenting stress over time is reported to affect the mental health of children with physical and mental disabilities (Baker et al., 2003). Therefore, attention to the parenting stress also in the context of reconstructive surgery is necessary when parents of children with LLI and CLP have to subdue their stress for many years of surgical treatments.

All psychological measures, apart from the ones developed for these studies, have been used extensively in process research on other health conditions in children and have been found to generate valid data. However, the small groups and varied diagnoses in our studies might limit generalization.

It is well accepted generally that psychopathology is not an associated feature for individuals with craniofacial anomalies (McGregor, 1990, Speltz and Richman, 1997). This was in accordance with the findings in Paper IV.

The CL/P group was compared to a standard sample and clinical group data by means of the BYI data in the manual. The CL/P group data on BYI had a profile closer to the standard sample than to the clinical group of patients with psychiatric diagnoses. We have no data on patients’ post-surgery health. But, in general, the CLP patients have undergone several surgical interventions up to the age of the examined group.

There were indications that one fourth of the CL/P group had problems, according to heightened scores on depression, anxiety and anti-social behav-
iour on the BYI. By using the YQ-CLP, the connection was evident between
the incidence of bullying and the report on the BYI Anger and Anxiety scale.

Psychological assessment is recommended since teasing is greater among
the CLP children. They are more depressed and anxious, have behaviour
problems and are unhappy with their facial appearance, according to a study
by Hunt et al. (2006).

The study by Herskind et al. (1993) where a higher suicide rate for CLP
affected individuals is reported, makes it important to take seriously the data
on mental health presented in this thesis; especially since mental health
might get worse by age. In the study by Cheung et al. (2007) adult CLP indi-
viduals reported a lower self-esteem and QOL.

Self-Esteem in the Three Patient Groups

Self-esteem was found to be above median level for the three patient groups.
Especially the CL/P patients’ self-reports were higher than expected. Many
recent studies have underlined the competence of parents and the cleft
teams’ engagement in their roles to support the children with CLP and other
disfigurements. The CL/P patients’ good self-esteem can be the result of
good awareness by people surrounding the child. However, a high self-
report might be an overestimation of one’s self and a kind of complacency
rather than having the capacity to construct a reviewing-of-self. The parents’
role of buffering the children’s self-esteem is very determining but the effect
of this support is not fully understood. More research is required in order to
know if there should be other components in the parenting process for opti-
mal support of the self-esteem and ability to cope with difficulties related to
the condition of a deformity (Cheung et al. 2007). Studies concerning Basic
and Earned self-esteem are interesting in this context of how self-esteem
develops (Johnson, 1997).

The new questionnaire of self-esteem YQ-CLP was in correspondence
with the BYI item of self-esteem. However, the new questionnaire contrib-
uted to information on a developmental effect of self-esteem and what had
diminished and what had been supportive for the sense of a good self-
esteeem. The study (Paper IV) could not answer the question if the self-
esteeem is either basic or multidimensional. By answering how they estimated
their self-esteem, indicated a global self-esteem while the answers to what
affected self-esteem was on a multidimensional level. However, the fact that
females with CL/P rated their self-esteem lower when growing older, could
imply the multidimensional impact of gender and cultural factors on self-
perception (Elbogen et al., 2001). Even then, in the theoretical debate on
self-esteem there is no consensus if a global or a multidimensional self-
esteeem is the most appropriate way to understand and measure self-esteem
(Lindwall, 2004).
The formulas need revisions both for validity, reliability reasons and factor analysis. Several items seemed to be of no interest or hard to remember. Patients and parents had, e.g., very few memories if there had been practical, social, psychological or medical problems in connection with surgery at different times. Remembering affects associated with surgery were also difficult.

The results of the new, specific condition formulas YQ-CLP and FI-CLP can be seen as the initial outcome for further testing of the psychometric qualities and testing the formulas on other groups of patients with deformities and defects in appearance.

Social Interaction

In Papers I and II the Activity level of the patients with LLI and PE were lower than in the control group. This was surprising, especially for the PE group that had a minor deviance in appearance.

The reconstructive surgery improved aspects of the children’s mental health such as less Attention problem and Somatic complaints (PE group) and also scores on Aggressive, Attention and Externalizing behaviour and self-rated depression and anxiety scores (LLI group). In future this effect could help these children to be comfortable in social situations.

Through the YQ-CLP and FI-CLP (Paper IV) we learned that the adolescents had, in general, found their way to satisfying social interaction with peers mostly by the support of parents, friends, sports and music (Broder, 2001). School played a less important role, which was somewhat unexpected. The lack of support reported by the adolescents could be an area for research in order to facilitate interaction at school and with peers.

The adolescent affected by CL/P were interested in group consultation as an intervention in the health care system of CLP. This is positive since studies have underlined the efficacy of support groups. A support group program for adolescent girls with the goal to improve self-esteem has proved to increase social support, body image, and self-efficacy (Steese, 2006). Social training programs also for children with craniofacial disfigurements have been successful in order to counteract bullying (Lovegrove and Rumsey, 2005). They declare that children with deformities need us to pay more attention to their behaviour. If there are signs of insecurity, shyness or avoidance behaviour, they need help to overcome these problems, otherwise their disability to cope might hinder their social interaction. Ignoring the impact of appearance and bullying will not make it stop. Rather, active social training and psychological support by others with similar conditions can be of good help (Lowegrove and Rumsey, 2005, Maddern et al., 2006).

Support to parents was also suggested by the adolescents but, interestingly enough, not family consultation. This could be a sign of the adoles-
cents’ desire to be independent and not willing to share too much with their parents. It could also be perceived as mental pain and a risk of emotional flooding and negative changes of the self if talking together with the parents (Orbach, 2003).

Methodological Considerations
Sample Size and Representativeness of the Sample
In *Paper I* the sample size of 27 LLI patients was small, but most of the clinical population of the paediatric orthopaedic patients during this period participated. Eight patients were excluded because of pre-operative problems not related to LLI; four were excluded because they missed the pre-operative psychological assessment; three had had their second or third lengthening and were therefore excluded. During the time of the study during 1997-2006, 42 patients with LLI were operated on.

The patients and their parents were invited to the assessment when they visited the surgeon. One might ponder the question, whether this affected their response to the study – the willingness to participate and how they responded to the psychological measures.

The child and parent were together during the interview but when the self-report/tests were administered the child was alone with the psychologist. The reason for having child and parent together during the first part of the assessment was trying to maximise the security for the child in the context of reconstructive surgery.

At the follow-up assessment one year after surgery the patient/parent reports on the satisfaction rate could be dependent on the relation to the surgeon. However, the psychological assessment, both before and after surgery, took place in another setting, at the Child and Adolescent Psychiatric Clinic, which hopefully gave place for an alternative conversation than did the medical setting. Another aspect of the psychological assessment was a semi-therapeutic situation where the patient and parent could talk about difficulties, but also reflect on how they had coped. The self-reports on psychological issues and self-esteem might also be fruitful for a reflective and resilient perspective on the situation (Rusch et al., 2000). Altogether, the risk for the subjects’ biased responses regarding satisfaction must be considered.

Always when using self-reports the scale scores should be interpreted with care and caution. From a study by Meijer et al (2008) we learn that using Harter’s self-perception profile and combined information from observation and interviews demonstrate that similar score profiles may result in a different interpretation. Children can have a less developed self-concept and/or problems with understanding the meaning of the questions.
In Paper II the participation was less satisfying. Out of 42 patients in the pre-surgery assessment, only 31 took part in the follow-up assessment one year after surgery. These PE patients were probably less dependant on the surgeon after the otoplasty intervention than were the LLI patients after the demanding leg-lengthening procedure. More frequently the PE patients and their parents found obstacles for taking part in the study. There was no medical examination except for a visit to the nurse one week after surgery. One year after surgery the PE patients had left the reconstructive surgery behind in different way than the LLI group. The PE patients were invited to the psychological assessment, with no connection to the medical ward. In that way the minor defect in appearance might have been less motivating for joining the psychological part of the study.

The follow-up time one year after surgery (Papers I and II) was decided out of the criteria of bone modelling for LLI patients – but a longer follow-up time might have changed the results in both directions. On the one hand, the improvement in behaviour reported by the parents could, for instance, have diminished when the event of the surgery was less focused on. On the other hand, improved function and appearance might enhance self-esteem after more time had passed.

In Paper III the participants were invited according to the waiting-list with a letter presenting the aim of the interview study. The study was described as an interactive interview with patients with CLP, parents, surgeon, and psychologist. All the 6 invited patients accepted to take part in the interview.

Following a systemic qualitative way of thinking, the study fulfilled its intention: to learn from a sample of adolescents with CLP about their self-esteem and its development over childhood and adolescent years.

The results of the interview study must also be considered with caution because of the small sample size of 6 patients. Therefore the themes that came out of the interviews were further explored in the main study (Paper IV) and they proved the importance of examining ideas and results in a larger group. For instance, the fathers in the preliminary study participated more and played a more active role than the mothers in relation to the CLP treatment situation. According to the fathers, the mothers were scared both of the operation theatre and of seeing blood. In the main study (Paper IV) this was not the case. On the contrary, as many as 47% of the fathers were reported by the mothers to have crises in connection with the birth of their child.

For all the studies the fact is that larger samples would have given more information and made the data analyses more reliable.

In Paper IV, 26 CL/P patients participated during the period of June 2005 to June 2008. Three patients (males) did not want to take part in the study. Two were missed out because of logistic reasons or because there was no psychologist on duty. Four patients (3 males and 1 female) did not want the
surgery when they were called in. There were no difference between participants and non-participants in any demographic variable.

One limitation of this population is the sample size, and the ratio male/female: 17/9. However the ratio 17/9 is consistent with the increased population of cleft lip in boys. The study also has its limitation by the fact of diverse CLP diagnoses which can imply different comorbidity factors for the CL/P group, for instance intellectual capacity (Persson et al., 2008). The close interplay of craniofacial and brain development might include genetic factors interacting with the individual’s capacity for social interaction (Mueller et al., 2007).

The Parents’ Presence During the Interview
This probably affected the reports of the adolescents and reflects the dependency between child and parent, a factor hard to exclude even if the adolescent is alone during the assessment. In the interview study (III) the criteria of a safe and normal situation was our priority, more so than a separate and possibly non-biased response situation for the young patient with CLP.

Memory issues
Children’s cognitive capacity as well as psychological understanding makes it difficult to remember and to answer retrospective questions. Take, for instance, the looking back on earlier episodes in the cleft treatment process or what the children remember of good and bad experiences at surgery events and their self-esteem at different ages. Children’s way of perceiving themselves and the world is egocentric, according to Piaget, which also affects their reflecting on the fact of living with CLP. They assess their mental health and self-esteem by their specific experience (Singer and Revenson, 1997).

The Strength of the Studies
The collaborative work with surgeons was accomplished in order to explore the effect of children’s mental health during the process of reconstructive surgery. Papers I and II are prospective studies on clinical groups that had not been studied before in a comparable way.

Interactive interviews with adolescents aiming at understanding the affect CLP can have on self-esteem and social interaction during childhood and adolescent years (Paper III). The new measures YQ-CLP and FI-CLP were explored in Paper IV, which also allow replication for future research in CLP samples and related fields.
Implication for Future Research

In this thesis the implication to develop a specific condition questionnaire in order to catch the specific difficulties which the children with deformities are confronting was focused on. The affected adolescents have been invited to interactive interviews and a questionnaire study with the goal better to understand the development of self-esteem. Triangulation of information was also addressed.

However, not addressed in this study were other important implications recommended for studies of CLP affected patients and patients with defects in appearance and function such as: multi centre studies in order to have homogeneous samples and long-term follow-up studies to see the effect of treatment interventions.

Study Designs

The prospective design in Papers I and II could not be applied because the patients in Paper III and IV were of a different age group. It seemed appropriate to study the CL/P patients in an age group when they were more cognitively and psychologically equipped to inform us on how they were affected by their condition during childhood and at present times. That was also the reason to choose other measures for mental health and self-esteem. Paper III was an interactive interview with adolescents with CLP. From the interview, current research data, and clinical practice the new measures YQ-CLP and FI-CLP were developed to examine the self-esteem and social interaction in CL/P affected adolescents.

Paper IV had a retrospective design with the aim to explore the new formulas. Using these new formulas created a clinical situation where patients and parents could answer the questions of the new formulas separately and in their own tempo, assisted by the psychologist.

The CL/P group in Paper IV was compared with a standard group and psychiatric samples by using the mean values presented in the Swedish manual of BYI (Tideman, 2004). Thus, the data should be considered as approximate.
Conclusions

- Reconstructive surgery by the Ilizarov technique does not pose a threat to the children’s mental health according to a one-year follow-up study (I).
- The effect of reconstructive surgery when there is a minor or major defect in appearance and function was demonstrated on both conditions. The LLI group gained more than the PE group in improved mental health (II).
- The interview study of adolescents with CL/P gave important themes on self-esteem and social support which resulted in two new formulas, Youth Questionnaire-CLP and Family Interaction-CLP (III).
- The adolescents with CL/P have a good self-esteem but the females in this group reported a diminished self-esteem from primary to secondary school age compared to males. Parents reported the opposite trend. A majority of the adolescents were bullied and their mental health was affected (IV).
Final Thoughts and Remarks

Self-esteem in the three patient groups demonstrates that children with Leg Length Inequalities (LLI) have, before surgery, the lowest level of self-esteem, and there is no significant change at the one-year follow-up.

Children with Prominent Ears (PE) have a self-esteem level somewhat higher and there is no effect on self-esteem measures after reconstructive surgery, according to ITIA.

The CL/P adolescents report an above median level of self-esteem earlier in life, also with a trend of no change, despite continuous reconstructive treatment, on the new formula YQ-CLP and on BYI. However, when studying the females in this group, there seems to be a strong effect when they grow older, since their self-esteem was reported to diminish in their teens, contrary to the males’ estimation of their self-esteem.

The parents report a positive development of self-esteem in their children on the FI-CLP. This demonstrates the complexity of the concept of self-esteem and the need for further studies. In literature there is still no consensus whether gender influences the satisfaction with CLP care (Turner et al., 1998).

Even though the studies did not point to clear effects of reconstructive surgery on self-esteem (Paper I and II), the condition-specific questionnaires involved valuable information in the search for self-esteem related questions. The child and adolescent patients with CLP encounter a range of appearance-related problems, such as being teased, feeling self-conscious, being upset about the reactions by those around and feeling depressed (Simis et al., 2001). Maybe this motivates the request for reconstructive surgery. It would be easier if the community could better understand that the request for surgery should not depend on high or low self-esteem, but rather on the functional aspects of surgery. In the meantime these findings have clear implications for the health-care services to provide targeted psychosocial treatments for the children and adolescents within the area of reconstructive plastic surgery.
Suggestions for the Use of the Self-Esteem and Social Interaction Measures YQ-CLP and FI-CLP

Self-esteem is a sensitive and important part of a young person’s mental health. Having a deformity might make it even more difficult to talk about one’s self-esteem.

The development of self-esteem includes the identification with emotionally important people and the process of finding important role models. Exchanging thoughts and feelings of how other people react on you and what effect your own behaviour has on them (Bachtin, 1981), might be strengthening for your well-being and self-esteem. And the image you have created of yourself can, finally, be fruitful to share with someone with similar life conditions.

Using the YQ-CLP in clinical practice in both individual and group settings with CLP affected adolescents is recommended, especially since it can be used in the activities suggested by the patients who participated in study IV.

FI-CLP was developed for individual meetings with parents of the adolescents with CL/P in study IV. The thesis has demonstrated that the parents do have much in common and appreciate meeting other parents in the healthcare treatment situations. The adolescents reported very competent parenting, but the parents themselves expressed loneliness and despair when struggling alone with a non-understanding environment.

However, in recent studies, as well as in these studies (Paper III and IV) the knowledge of the importance of meeting other parents and children in hospital has been encouraging.

The FI-CLP formula would be informative in clinical practice in order to help parents express experiences and the need of support.
Barns psykiska hälsa i samband med rekonstruktiv kirurgi- studier av barns självbild och sociala interaktionsförmåga

Målet med avhandlingen var att undersöka hur barn reagerar psykologiskt i samband med olika typer av rekonstruktiv kirurgi. Den rekonstruktiva kirurgin, som syftar till att normalisera funktion och utseende, har varit framgångsrik, men vilken effekt kirurgin har på barnens psykiska hälsa, självbild och sociala förmåga är mindre utforskat. Föräldrarna har också deltagit i undersökningen genom att skatta sina barns psykiska hälsa, självbild och sociala förmåga samt egna reaktioner under den rekonstruktiva kirurgiprocessten.

Tre patientgrupper har studerats för att utröna de svårigheter dessa barn och ungdomar är utsatta för genom sin avvikelse i utseende och funktion.

- Barn med benlängdsskillnader
- Barn med utstående öron
- Ungdomar med läpp-käk- och gomspalt (LKG)

Frågeställningar

- Att undersöka barnens psykiska hälsa före och efter benförlängningsbehandling.
- Att fastställa eventuella skillnader i effekt av rekonstruktiv kirurgi på psykisk hälsa och självbild när barnet har en mindre defekt i utseende och funktion, som vid utstående öron jämfört med barn med benlängdsskillnader.
- Att utforska socialt stöd och självbild hos ungdomar med LKG i samband med att de genomgår en korrektion av läpp och näsa.
- Att utforska självbildens och den sociala interaktionsförmågan med ett frågeformulär som är speciellt utvecklat för ungdomar med LKG.
Resultat

I de två första studierna konstaterades att benförlängningspatienterna hade sämre hälsa och självbild än en kontrollgrupp. Ett år efter den rekonstruktiva kirurgen förbättrades båda patientgruppens (benförlängnings- och öronplastikpatienter) psykiska hälsa, men självbilden förbättrades inte. Detta gjorde att självbildsbegreppet stod i fokus för den fortsatta forskningen.

I den tredje studien som bestod av en interaktiv intervju med 6 ungdomar med läpp-käk- och gomspalt (LKG) framkom intressanta synpunkter på hur de uppfattade sin självbild och hur de hanterade svårigheter. Resultaten användes till att skapa två frågeformulär för ungdomar med LKG och deras föräldrar.

I den fjärde studien kunde vi med hjälp av de nya frågeformulären konstatera att ungdomarna hade ett bra stöd från sina närmaste och en bra självbild, men att flickornas självbildsskattning sjönk jämfört med pojkarnas i åldern 10-19 år. Den psykiska hälsan för LKG-gruppen är nära normen för ungdomar i allmänhet, men riskfaktorn mobbning gör att denna patientgrupp ändå bör få ökad uppmärksamhet inom BUP och i de speciella ”spaltteam” som finns på Plastikkirurgiska kliniker.
Appendix I

Name:
Date:

In order of your CL/P operations, which one will this be? ……..

Youth Questions, Cleft Lip/Palate (CL/P)

1. When did you understand that you looked different, were different from other children?
   At what age? …………………………………………………………....

2. Did you get a lot of attention because of your CL/P?
   a. A lot
   b. Some
   c. A little
   d. None

3. How often do you think of your CL/P?
   a. Always
   b. Often
   c. Sometimes
   d. Rarely
   e. Never

4. If “never”, when did you stop thinking of your CL/P?
   …………………………………………………………………………

5. When did you stop seeing your CL/P as a handicap?
   …………………………………………………………………………

6. How much did you talk about CL/P at home?
   a. A lot
   b. A little
   c. Not at all
7. At home, how much have you been talking about operations and treatment results?
   a. A lot
   b. A little
   c. Not at all

8. At home, who was the easiest person to speak with?
   a. Mum
   b. Dad
   c. Both
   d. Somebody else

9. Who used to accompany you to the hospital for your operations?
   a. Mum
   b. Dad
   c. Both
   d. Somebody else
   Who would you rather have liked to be in your company?
   ......................................................................................................................

10. At what age were you allowed to take part in deciding upon a possible operation?
    6 years  8-12 years  13-19 years

11. What was your experience of the speech improving operation at preschool age?
    a. No improvement
    b. Some improvement
    c. Great improvement
    Please, describe in your own words:
    ......................................................................................................................
    ......................................................................................................................
    ......................................................................................................................

12. What was your experience of the appearance improving operation of lip/nose during compulsory school years?
    a. No improvement
    b. Some improvement
    c. Great improvement
    Please, describe in your own words:
    ......................................................................................................................
    ......................................................................................................................
    ......................................................................................................................
13. What was your experience of the appearance improving operation of lip/nose during secondary school years?
   a. No improvement
   b. Some improvement
   c. Great improvement
   Please, describe in your own words:
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………

14. Did bullying of any kind occur?
   a. A lot
   b. A little
   c. None
   If so, when did it happen?
   ……………………………………………………………………………
   How?
   ……………………………………………………………………………
   ……………………………………………………………………………

15. Here follows a list of emotional reactions:
   a. Feeling of dissociation
   b. Anger
   c. Distress/despair
   d. Shame
   e. Fault
   f. Curiosity/interest
   g. Tenderness
   h. Love
   i. Shock ………………………………………..

At different ages, what emotional reactions did you feel after your operation (which you yourself remember or which your parents have told you about)?
Please, write down an emotion from the above list of emotions.

18 months-3 years 4-7 years 8-12 years 13-19 years
………………………….. ………….. …………… ……………
………………………….. ………….. …………… ……………
………………………….. ………….. …………… ……………

Please, tick off:
– I remember – I remember – I remember – I remember
– parent told me – parent told me – parent told me – parent told me
16. Do you perceive yourself as having been socially isolated (staying at home more often, mostly seeing your school-mates and not so many others of your age) on account of your CLP?
   a. A lot
   b. Somewhat
   c. A little
   d. Not at all
   e. Please, describe it in your own words (like staying at home, e.g., by the computer or the piano, and it has nothing to do with your CLP):
      ……………………………………………………………………………
      ……………………………………………………………………………
      ……………………………………………………………………………

17. Who or what has for you been of great help for managing your CL/P?
   a. Parental support
   b. The work of the school staff
   c. The work of the hospital staff
   d. Friends
   e. Sports or other activities or clubs
   f. Church
   g. Music, art, or other creative activity

18. What was your self-esteem like before and what is your self-esteem like now on a scale between 1 and 9, where 1 is the lowest possible self-esteem (discontented with everything in yourself) and 9 is the highest self-esteem (content with everything) at the ages of:
   6 years 8-10 years 13-19 years
   …………… …………… ……………

How come that you did feel and do feel that self-esteem? Please, describe in your own words:
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………

19. What made your self-esteem/your view of yourself waver?
   a. School reports
   b. Meeting new grown-ups
   c. Meeting new people of your own age
   d. Operations and treatments
   e. Nothing in particular
   f. Other things:
   ……………………………………………………………………………
20. If you were to be the parent of a child with CL/P, what feelings would you have?
   a. Feeling of dissociation
   b. Anger
   c. Distress/despair
   d. Shame
   e. Fault
   f. Curiosity/interest
   g. Tenderness
   h. Love
   i. Shock

21. Would your feelings be different if you had a girl or a boy?

   Yes    No

   If so, what would your feelings be like?

22. What help do you think that you as a parent would like to get?

23. If you were able to change something on or for yourself, what would it be? Tick off and place in order of preference, i.e., write 1 for the thing you would like the most to change.
   a. …Talent
   b. …Looks/face
   c. …Body weight
   d. …Body shape
   e. …Social ability
   f. …Economic situation
   g. …Place to live
   h. …Relations to comrades
   i. …Family affairs
24. What kind of help would you like to get as a patient at the Plastic Surgery Clinic?
   a. Group discussions for young people with CL/P, including information and free talking
   b. Group proficiency practice for young people with CL/P, where everybody would practise together both on how to get into contact with new people and to appear in different social connections
   c. Individual talks with a psychologist
   d. Parental support
   e. Family talks
   f. Other things:
   
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………
   ……………………………………………………………………………

Below, you may write down your views on the questionnaire or whatever else you have come to think of while answering the questions.

…………………………………………………………………………
…………………………………………………………………………
…………………………………………………………………………
…………………………………………………………………………
…………………………………………………………………………
…………………………………………………………………………

Thank you for your participation!
Appendix II

Father: Please, fill in your own experiences and what
Date: you believe the child’s other parent reacted
on and how she/he handled the situation.

Family Interaction, Cleft Lip/Palate (CL/P)

Background data, socio-economic factors
1. Child’s sex B G
2. Child’s number in circle of sisters and brothers ..............
3. Parents’ professions, education Mother: ......................
   Father: .......................:
4. Civil status ........................................
5. Nationality ...........................................
6. Country and town ........................................

The child’s diagnosis and condition
1. Type and extent of deformity ........................................
   Family occurrence of CL/P ........................................

2. Father’s age at delivery .................................
   His first child? Yes No If not, what number of child? .......

3. What was your very first feeling when you saw your child for the first time?
   A Feeling of dissociation
   B Anger
   C Distress/despair
   D Shame
   E Fault
   F Curiosity/interest
   G Tenderness
   H Love
   I Shock When did the shock pass off? ..............
   What help did you get? .........................
4. What feeling (see question 3) was predominant in the relation to your child at the following ages?

**For the father:**
0-2 months .................................................................
3-9 months .................................................................
10-18 months .............................................................
3 years ......................................................................
6 years .....................................................................
7-9 years .................................................................
10-12 years ............................................................... 
13-16 years ............................................................... 
17-19 years ............................................................... 

5. If you had had a child of the opposite sex, would it have changed your reaction?
Yes  No

If so, how?
......................................................................................
......................................................................................

6. Mother’s age at delivery .........................
Her first child?
Yes  No  If not, which one in the order of children? ........

7. What was her first feeling when she saw her child for the first time?
A  Feeling of dissociation
B  Anger
C  Distress/despair
D  Shame
E  Fault
F  Curiosity/interest
G  Tenderness
H  Love
I  Shock  When did the shock pass off? ..............
What help did you get? ......................

8. If she had had a child of the opposite sex, would it have changed her reaction?
Yes  No
9. What feeling (see question 3) was predominant in the relation to her child at the following ages?

For the mother:
0-2 months .................................................................
3-9 months .................................................................
10-18 months .........................................................
3 years .................................................................
6 years .................................................................
7-9 years .................................................................
10-12 years ...............................................................
13-16 years ............................................................... 
17-19 years .................................................................

10. For the father: Were your feelings different compared to those towards siblings, if any, without CL/P?
Yes  No

11. Do you believe that your contact with the child became different on account of her/his CLP?

Please, describe in your own words:
................................................................................
................................................................................

Research has found that parents have sometimes reacted by not being able to take to the child, alternatively have come to overprotect the child on account of her/his CLP.

Tick off on the scale below, if you find that you can recognise the reaction.

The very first week, 0-1 week:

Did not take to child..............................................................overprotective

1 week-6 months:

Did not take to child..............................................................overprotective
12. Do you believe that the contact with your child is different on account of her/his CL/P?

Please, describe in your own words:

……………………………………………………………………
……………………………………………………………………

Tick off on the scale below, how you perceive the contact with your child to be now:
Not at all overprotective........................................ Very much overprotective

13. What has been your attitude to the reactions of people around you concerning your CL/P child?
   A  Openly informative
   B  Avoiding
   C  Anger

14. Do you consider your child to have become socially isolated on account of her/his CL/P?
   A  Very much
   B  Somewhat
   C  A little
   D  Not at all
   E  Please, describe in your own words (e.g., likes being at home alone; for example, by the computer or the piano and it has nothing to do with CLP):

   …………………………………………………………………
   …………………………………………………………………
   …………………………………………………………………

15. What were your hopes at the prospect of the present CL/P operation?
   A  Expectant
   B  Neutral
   C  Despondent

16. Were there obstacles to the operation?
   A  Child’s attitude
   B  Parents’ attitude
17. At different ages, were there problems before the operation?

<table>
<thead>
<tr>
<th>Plast Surg: lip</th>
<th>Soft palate closure of cleft rest</th>
<th>Speech improving correction</th>
<th>Wrong jaw operation</th>
<th>lip/nose</th>
</tr>
</thead>
<tbody>
<tr>
<td>3-5 months</td>
<td>6-17 month</td>
<td>18 m.-3 y.</td>
<td>4-7 y.</td>
<td>8-12 y.</td>
</tr>
</tbody>
</table>

Please, describe the problems in your own words:
……………………………………………………………………………………
……………………………………………………………………………………
……………………………………………………………………………………

18. How did you decide on an operation at the different ages – doctor’s recommendation, parents’ or relatives’ or child’s wish/conduct?

<table>
<thead>
<tr>
<th>3-5 months</th>
<th>6-17 months</th>
<th>18 m.-3 y.</th>
<th>4-7 years</th>
<th>8-12 years</th>
<th>13-19 years</th>
</tr>
</thead>
</table>

19. At what age was your child allowed to take part in deciding on possible operation?

<table>
<thead>
<tr>
<th>18 months-3 years</th>
<th>4-7 years</th>
<th>8-12 years</th>
<th>13-19 years</th>
</tr>
</thead>
</table>

20. Were you able to talk with the doctor after the operations?

a. A lot
b. A little
c. Not at all

21. Were you able to talk with the nurse after the operations?

a. A lot
b. A little
c. Not at all
22. Did you experience problems after the operations?

<table>
<thead>
<tr>
<th>Age Group</th>
<th>3-5 months</th>
<th>6-17 month</th>
<th>18 m.-3 y.</th>
<th>4-7 y.</th>
<th>8-12 y.</th>
<th>13-19 y.</th>
</tr>
</thead>
</table>

Please, describe the problems in your own words:

………………………………………………………………………
………………………………………………………………………
………………………………………………………………………

23. At home, do you talk about the operations and the result of the treatments?
   a. A lot
   b. A little
   c. Not at all

24. Is any one parent sensitive to blood, hospital surroundings or the like?
   Mother:
   a. A lot
   b. A little
   c. Not at all

   Father:
   a. A lot
   b. A little
   c. Not at all

   Both:
   a. A lot
   b. A little
   c. Not at all

25. Do you still see your child as handicapped?
   Yes  No

   If no, **when** did you stop seeing your child as handicapped?
   …………………………………………………………………………
   …………………………………………………………………………

55
26. Have you felt the need to support your child’s self-esteem?
   a. Very often
   b. Often
   c. Rarely
   d. Never

27. What has the mother done to support her child?
   a. Moral support (e.g., you are good the way you are; the value of a person lies in their inner qualities more than their looks)
   b. Informatively (e.g., she has told her child and the school about CLP, treatments, what it will be like)
   c. Socially (supported the child to join in various activities and to meet youngsters of the same age)
   d. Put her trust in the medical services (said that the doctor can explain or that operations will make it better)
   e. Other things ………………………………………………………

28. What have you done to support your child?
   a. Moral support (e.g., you are good the way you are; the value of a person lies in their inner qualities more than their looks)
   b. Informatively (e.g., you have told your child and the school about CLP, treatments, what it will be like)
   c. Socially (supported the child to join in various activities and to meet youngsters of the same age)
   d. Put his trust in the medical services (said that the doctor can explain or that operations will make it better)
   e. Other things ………………………………………………………

29. If you were to estimate your child’s self-esteem on a scale between 1 and 9, where 1 is the lowest self-estimation (dissatisfied with everything in themselves) and 9 is the highest self-estimation (satisfied with everything in themselves) – when was the self-esteem at its lowest? Age? …

   1 2 3 4 5 6 7 8 9

   1 2 3 4 5 6 7 8 9

   Now? Age? …

   How come that your child did and does have that self-esteem? Please, describe in your own words:
   …………………………………………………………………………
   …………………………………………………………………………
   …………………………………………………………………………
Below, you may write down your views on the questionnaire or something else that you have come to think of when answering the questions:

........................................................................................................................................
........................................................................................................................................
........................................................................................................................................
........................................................................................................................................
........................................................................................................................................
........................................................................................................................................

Thank you for your participation!
To Viveka Sundelin Wahlsten, my first supervisor, for never-ending energy and enthusiasm that helped me during my journey to accomplish this thesis.

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