Table of contents

Acknowledgements

Summary

1. Introduction ........................................................................................................................................... 1
   1.1. Background ................................................................................................................................. 1
   1.2. Cleft lip and/or palate ................................................................................................................ 2
   1.3. Consequences and treatment of cleft lip and/or palate .......................................................... 3

2. Growing up with a cleft: Psychological functioning ................................................................. 4
   2.1. Theoretical and empirical perspectives ....................................................................................... 4
       2.1.1. A cognitive–behavioural perspective on facial appearance ................................................. 4
       2.1.2. Visible differences and appearance evaluations .................................................................. 6
       2.1.3. Self-perceptions .................................................................................................................. 7
       2.1.4. Psychosocial experiences and friendships ......................................................................... 8
       2.1.5. Emotional adjustment ........................................................................................................ 10
       2.1.6. Developmental patterns ...................................................................................................... 11
       2.1.7. Risk and protective factors ................................................................................................ 12
       2.1.8. Cognitive functioning ......................................................................................................... 13
       2.1.9. Summary ............................................................................................................................ 14
   2.2. The presence of associated conditions ...................................................................................... 14
       2.2.1. Traditional exclusion criteria in cleft research ................................................................. 15
       2.2.2. Summary ............................................................................................................................ 19
   2.3. Limitations in the research field ............................................................................................... 19

3. Aims and research questions ........................................................................................................... 20

4. Methods ............................................................................................................................................ 21
   4.1. Procedure and research ethics .................................................................................................... 21
   4.2. Participants ................................................................................................................................... 22
       4.2.1. The comparison group in Paper 2 ....................................................................................... 23
       4.2.2. Prevalence of associated difficulties in the cleft samples ................................................ 23
       4.2.3. Ethnicity ............................................................................................................................. 26
       4.2.4. Attrition .............................................................................................................................. 26
   4.3. Measures and instruments ......................................................................................................... 27
       4.3.1. Dependent and independent variables in Paper 1, 2, and 3 ............................................... 27
       4.3.2. Visibility of cleft ............................................................................................................... 27
       4.3.3. Presence or absence of an associated difficulty ............................................................... 27
       4.3.4. Psychosocial experiences, social acceptance, and close friendships .............................. 28
       4.3.5. Satisfaction with appearance ............................................................................................. 29
       4.3.6. Cognitive and psychological functioning .......................................................................... 30
       4.3.7. Emotional adjustment ......................................................................................................... 31
   4.4. Statistical analyses ..................................................................................................................... 31
       4.4.1. Testing of mediation and moderation ................................................................................ 32
       4.4.2. Handling of missing data .................................................................................................. 33
5. Results .................................................................................................................................34
   5.1. Results in Paper 1 ........................................................................................................34
   5.2. Results in Paper 2 ........................................................................................................35
   5.3. Results in Paper 3 ........................................................................................................36

6. Discussion ..........................................................................................................................36
   6.1. Methodological issues .................................................................................................38
   6.1.1. Measurement issues ..............................................................................................38
   6.1.2. Lack of control group ............................................................................................40
   6.1.3. The issue of causality ............................................................................................41
   6.1.4. Representativeness and generalizability of the sample .........................................42
   6.2. The presence of associated difficulties .......................................................................44
   6.3. Adolescents with a visible versus a non-visible cleft ..................................................45
   6.4. Psychosocial experiences, friendships and social acceptance ....................................46
   6.5. Social cognitions and attributions .............................................................................49
   6.6. Gender differences .....................................................................................................50
   6.7. Resilience or absence of risk? ....................................................................................51
   6.8. The importance of appearance evaluations ..................................................................53
   6.9. Clinical implications: Treatment and prevention .......................................................54

7. Conclusion ............................................................................................................................56

REFERENCES ..........................................................................................................................58

Appendix

List of tables

Table 1 Overview of empirical studies investigating psychological functioning in children and adolescents with a cleft. ................................................................. 16
Table 2. Distribution of participants across the three papers .................................................24
Table 3. Frequency of associated difficulties in the child and the adolescent sample. ........25
Table 4. Frequency of associated difficulties in children and adolescents with a visible and with a non-visible cleft. ................................................................. 25

List of figures

Figure 1. Schematic presentation of: a) Unilateral cleft lip alveolus, b) Complete unilateral cleft lip and palate, c) Complete bilateral cleft lip and palate, and d) Cleft palate only. .......3

Papers 1 - 3
Acknowledgements

This work would not have been possible without several people that have supported me during the three years I have been working with this PhD dissertation.

First, I want to thank my supervisors, Professor Anne-Inger Helmen Borge and Professor Nichola Rumsey. Thank you both for sharing your competence and experience, while always encouraging me along the way. In spite of a heavy workload, you have both been accessible to offer your expertise and your advice, which was very helpful during the ups and downs of this work. Thank you, Anne Inger, for sharing your optimism and enthusiasm, and for your valuable and important focus on friendships and resilience. Thank you, Nicky, for your faith in me, your warmth, and your invaluable competence in the field of research about visible differences. I am also greatly indebted to Associate Professor Ingela Lundin Kvalem. Thank you for sharing your competence and research ideas even before financial support for this work became a reality. Thanks for providing access to the data from the comparison group for the second paper, thus contributing to new perspectives in the interpretation of results. And thank you for your important suggestions and the time you generously spent helping me during the last week of work! I also want to thank the Norwegian Social Research (NOVA) for granting permission to use data from Young in Norway 2002. I am very grateful for help provided by Dag Erik Eilertsen with statistics for the second paper. Thank you to Donna Stevens who transformed this dissertation into readable English.

I worked on this study while in the employment of Bredtvet Resource Centre and the Oslo CLP Team. Being part of something bigger and on track while sitting in front of my computer and SPSS-files was quite valuable to me. Thanks to Anne-Berit and all colleagues at Bredtvet and Rikshospitalet University Hospital for giving me a feeling of belonging. A special thanks to my colleagues who were in charge of the clinical work as psychologists in the team, providing data for the study in spite of the extra work involved. Without you, Frøydis Tevik, Yvonne Severinsen, and Stine Meløy, I would easily have felt more lonely and vulnerable.

Some people have not been directly involved in this study but have nonetheless, by virtue of what they mean to me, provided me with motivation for my work. My thanks to both of you, Professor Frank Åbyholm and Professor Gunvor Semb, for your almost life-long true and absolute dedication to the treatment of children, adolescents, and adults born with a cleft lip and/or palate.
and their families, a dedication which has been a strong incentive for my own work as a clinical psychologist and researcher.

I also would like to thank friends and colleagues whose presence and trust in me provided motivation, support, closeness, and intimacy. I especially would like to thank all members of V&V (Karin, Kari, Ingvild, Kristine, Turid, and Brita), Hilda, Kirsten, Susan, Daniele, and Areana for friendship and encouragements all the way, my exercise partners Agnieszka and Grete, as well as Hanne-Marit for her patient listening skills. Thank you Line for your Word-expertise! Thank you, Kirsten and Jarl, for your warm presence. Thanks to my family and all other friends and colleagues, whom I cannot mention as, luckily, their names would fill this page! Thank you to my mother for contributing to this work by occasionally taking one of our children to the theatre, the forest, or home from school. Thanks to my father who housed me during two weeks in St. Laurent-la-Vernède, and gave me the extraordinary opportunity to concentrate totally on the dissertation, along with fantastic meals and French wines!

The biggest hug however, goes to the ones whom I love most of all, Samuel, Miriam, Anaëlle, and my husband, Magne. Thanks to you for reminding me of what means the most in my life – you! – and for accepting the fact that I have been less emotionally available than my conscience dictated during this long three-year period!

This work was totally financed by the EXTRA funds from the Norwegian Foundation for Health and Rehabilitation (grant 2005/2/0201), and supported by the parents’ organization, the Cleft-Lip and Palate Association (Leppe-ganespalteforeningen). I want to thank them for their support and faith in the project. I further wish to thank the Department of Psychology at the University of Oslo for the opportunity of participating in their PhD-programme.

Last, but not least, I want to thank all the children and adolescents, and their families who participated in this study, and in doing so, contributed with their thoughts, experiences, and strategies for coping with the challenges they have encountered. Without you, this work would have been worthless.

Kristin Billaud Feragen, Oslo, June 2009
Psychosocial adjustment in children and adolescents with cleft lip and/or palate:
Exploring risk and protective factors

Kristin Billaud Feragen
Bredtvet Resource Centre

Department of Psychology
Faculty of Social Sciences
University of Oslo
2009
List of papers

Paper 1


Paper 2


Paper 3

SUMMARY

Children and adolescents with cleft lip and palate and their families face challenges that are associated with this condition and its treatment. In addition to the burden of care related to surgical and orthodontic treatment, children and adolescents may also experience problems related to appearance satisfaction and social interaction. In spite of the recognition of the importance of subjective measures of appearance, little is known about protective factors at work in the associations between social experiences and appearance evaluations when there is a visible difference of the face.

This dissertation explores risk and protective factors in children and adolescents born with a cleft, based on cross-sectional data from a total of 661 children and adolescents, and 578 parents. Psychological assessment was done at age 10 for the child group, and at age 16 for the adolescent group. A semi-structured interview was used for the children, while the adolescents and the parents were asked to complete a questionnaire. This study includes measures of emotional functioning, perceptions of friendships and social acceptance, psychosocial experiences, and subjective measures of appearance.

The key findings were that whether the cleft is objectively visible in the face or not did not contribute to the explanation of the variation in psychosocial experiences or emotional well-being. In contrast, subjective measures of appearance were associated with psychological functioning. Secondly, positive psychosocial adjustment was associated with adequate emotional functioning, high satisfaction with appearance, and a lower frequency of reported teasing. Thirdly, subjective perceptions of appearance mediated the association between social acceptance and depressive symptoms. Fourth, adolescent boys with a visible cleft reported significantly more positive perceptions of friendships and social acceptance, and a lower occurrence of depressive symptoms than boys with no facial difference, findings that were interpreted as an indication of emotional resilience. Finally, gender moderated the adolescents’ satisfaction with appearance, girls with a visible cleft being less satisfied than girls with a non-visible cleft, or than the adolescent boys. However the interaction between cleft visibility and gender disappeared when information was included about peer harassment, thereby emphasizing the impact of social experiences on self-perceptions. Parental reports of teasing were not associated with psychosocial adjustment or the child’s satisfaction with appearance.
The results emphasize the subjective rating of appearance both as a potential risk and as a protective factor. Appearance satisfaction may be enhanced by positive social experiences, while dissatisfaction may be exacerbated by negative experiences such as harassment. The results thus support the notion of positive friendship experiences as protective factors in children and adolescents with a cleft lip and/or palate. The fact that appearance mediated the association between social acceptance and emotional adjustment regardless of whether there was a facial difference or not therefore adds to the existing literature on visible differences by demonstrating ordinary coping mechanisms in children and adolescents with a cleft.

The expected higher frequency of associated difficulties in children and adolescents with a cleft palate or submucous cleft palate than in cases of cleft lip and palate or cleft lip alveolus was confirmed. This study utilized the approach of carefully documenting associated conditions, in order to provide a more informed and inclusive approach than had been used previously. The recording of associated conditions made it possible to control for their potential effect on measures of psychological functioning. Existing literature has demonstrated a heightened risk for specific developmental problems and learning difficulties in children with cleft palate. However, by carefully registering the presence of associated difficulties in the sample, the results showed normal psychological adjustment or cognitive function in children with a cleft without an additional difficulty. The results also showed no differences between the two cleft types when children with associated difficulties were excluded.

This study addressed several important aspects in coping with a visible difference such as a cleft lip and palate. The following findings are emphasized:

- The significance of subjective ratings of appearance in contrast to objective diagnostic measures when assessing adjustment to a facial difference
- The importance of the associations between social experiences and subjective appearance satisfaction: the protective effect of friendships and social acceptance, and the risk factor related to peer harassment
- The need of assessing the overall emotional adjustment in the child
- The importance of careful registration of all associated conditions in order to be able to discern psychological mechanisms related specifically to the cleft condition, in contrast to mechanisms related to the presence of any additional difficulty.
1. Introduction

1.1. Background

In the 1960s, there was a book published entitled *The Scar*, by Bruce Lowery (1961). It was the history of the dramatic consequences in a young child’s life caused by the suffering of feeling visibly different. Jeff, a 13-year-old boy with a cleft lip and palate, did not understand the origins of his scar, so he was unprepared for dealing with other children’s reactions to his visible difference. Being unable to talk about his inner suffering, Jeff chose to reject those who approached him, as he so often felt rejected himself. Jeff was a vulnerable boy, trapped by his inability to protect himself and unable to cope with the negative social experiences that were part of his life’s story.

This is a child one meets in clinical work as a psychologist. The stigma of looking different may be compounded by the medical stressors of treatment. However, in spite of risk factors, what rapidly becomes evident in meeting children and adolescents born with a cleft lip and/or palate is the presence of protective factors and patterns of resilience. It also becomes evident that a lack of observable psychopathology does not necessarily mean easy adjustment. The question “What makes a positive difference?” comes up again and again in those meetings with children and adolescents and was a theme throughout the work for this dissertation. What are the factors that explain why some children strive and experience their cleft condition as a barrier to positive experiences, while others deal effectively with the challenge, and/or do not feel different or marked by their visible difference? The search for answers to these questions was the foundation of this study.

Much of the existing literature on facial differences has a pathological and problem-focused research approach. However, knowledge about protective factors may contribute to a better prevention of risk and psychosocial maladjustment. In spite of the clinical importance of the resilience perspective, empirically-based studies focusing on protective factors are still sparse within this area of research. In addition, many studies on the impact of visible differences are limited by methodological problems. First, most studies within the research area are limited by small sample sizes. A review paper (Hunt, Burden, Hepper, & Johnston, 2005) reports that among the articles included on the psychosocial impact of a cleft lip and palate, only five papers had more than 200 participants. The second problem is a lack of representative samples, as a result of non-centralized treatment settings. Furthermore, a large number of studies are based on samples with widely differing age groups, in order to recruit large
enough samples. Consequently, the validity of previous results as well as the ability to generalize them can sometimes be questionable. In addition, there is a lack of knowledge concerning differences between boys and girls in their adjustment to a visible difference such as a cleft, again due to small sample size, impeding comparisons across gender. Finally, the presence of other conditions in addition to a cleft lip and palate constitutes a challenge for the accurate measurement of psychological adjustment. Hence, it was imperative in this study to capitalize on the unusually large data set derived from clinical routine psychological assessments of children and adolescents in Norway, in order to explore risk and protective factors across gender, concerning psychosocial adjustment, subjective appearance satisfaction, and emotional well-being of individuals born with a cleft.

1.2. Cleft lip and/or palate

The embryologic development of the face takes place between the 5th and 9th week of gestation, while the development of the palate is completed two weeks later (Watson, 2002). Anything that interferes with the processes of cell fusion during facial development may result in a cleft. The type of cleft is dependent of the degree and the timing of this disturbance. The causes of this failure of neural crest cells to migrate properly and fuse are still not completely understood, and are multi-factorial in origin, involving interactions of both genetic and environmental factors (Jugessur & Murray, 2005).

Although cleft lip and/or palate occurs most frequently as an isolated anomaly, it is well known that it can also be associated with other congenital anomalies, and may constitute one of the many features of a syndrome (Lees, 2002). In Norway, approximately 120 children annually are born with a cleft in the lip and/or palate, a prevalence that corresponds to the documented occurrence of 1–2 per 1000 newborns (Abyholm, 1978; Sivertsen et al., 2008).

There is general agreement in classifying clefts into three main groups:

a) Cleft lip/alveolus (CLA), unilateral (UCLA) or bilateral (BCLA)
b) Cleft lip and palate (CLP), unilateral (UCLP) or bilateral (BCLP)
c) Cleft palate (CP) or submucous cleft palate (SMCP)\(^1\)

\(^1\) SMCP is an incomplete cleft of the palate, which very often remains undiagnosed unless the child develops defective speech as a result of the hidden cleft. Consequently, the number of recorded cases with SMCP does usually not correspond to its true prevalence in a population.
The cleft may vary in degree, and is usually described as an incomplete or complete cleft. All
degrees of severity are possible within the various cleft groups, and there are often wide
variations in tissue deficiency and configuration within each cleft group (Watson, 2002).
Some examples are presented in Figure 1 a-d (Shaw & Semb, 1993).

![Figure 1. Schematic presentation of: a) Unilateral cleft lip alveolus, b) Complete unilateral cleft lip and palate, c) Complete bilateral cleft lip and palate, and d) Cleft palate only.](image)

### 1.3. Consequences and treatment of cleft lip and/or palate

Norway utilizes a centralized model of treatment of cleft lip and/or palate, with two regional
teams (Oslo and Bergen), providing care for the entire cleft population. The Oslo team is
responsible for 2/3 of the Norwegian cleft population, treating approximately 70-80 babies
born annually with a cleft. This model of centralized treatment provides researchers with
access to large clinical samples that are representative of the study population. Treatment is
multidisciplinary, involving plastic surgeons, specialist nurses, orthodontists, speech
therapists, ENT-specialists, and psychologists. Other specialties may be involved when
necessary, such as geneticists and maxillofacial surgeons. The treatment protocol of the Oslo
team is presented in a chart in the Appendix. As can be seen from that chart, the treatment of
cleft lip and palate is not completed until the individual reaches adulthood.

A cleft lip and/or palate have potential implications for facial appearance, dental
development, hearing and speech, depending of type of cleft. Children with a cleft lip and/or
palate have to undergo several surgical and orthodontic interventions. As the child grows
older, speech may be deviant in terms of articulation or resonance. The child with a cleft lip
and/or palate may consequently feel visibly and/or audibly different from his/her peers. Even
if surgical treatment dramatically improves the baby’s initial appearance, the surgical scar will remain. This mark is located in the nose-mouth area, the part of the face that is the primary focus of attention in face-to-face interactions (Mertens, Siegmund, & Grüsser, 1993).

2. Growing up with a cleft: Psychological functioning

2.1. Theoretical and empirical perspectives

Much of the existing studies in this area of research have been based on two assumptions (Rumsey & Harcourt, 2005, p.29). The first assumption is that individuals with an objective, visible disfigurement differ from individuals without a visible disfigurement in the concerns about appearance and the processes involved in such concerns. Secondly, that negative experiences are presumed and expected in the presence of a disfigurement. Such experiences are thought to be directly related to the degree of visible difference. These suppositions have contributed to the development of a pathological and problem-focused approach to research in this area.

This study explored risk and protective factors in adjusting to a visible difference, based on a developmental and cognitive-behavioural perspective. Concepts are drawn from research fields such as health psychology, social psychology, and the psychology of appearance and body image. Within this developmental framework, the study explored how adjustment to a visible difference might be associated with positive or negative social experiences and interactions, perceptions of appearance, and emotional well-being, while also investigating the role of gender, age, and the presence or absence of other difficulties in addition to the cleft.

2.1.1. A cognitive–behavioural perspective on facial appearance

Thomas F. Cash developed a cognitive-behavioural model of body image development (Cash, 2002). This model distinguishes between developmental and concurrent factors that interact in the formation of a multidimensional body image. Historical or developmental factors refer to influences that may affect cognitive, emotional, and behavioural aspects related to our appearance. Four factors are proposed: cultural socialization, interpersonal experiences, physical characteristics, and personality attributes. These factors are believed to be in constant interaction with the fundamental body image schemas, which consist of two fundamental attitudinal elements: body image investment (importance of appearance), and body image
evaluation (self-appraisals of appearance). Both elements include affective, cognitive, and behavioural components. Hence, individuals who evaluate their own facial difference as having a negative effect on their appearance, and who place importance on the role of appearance will be more at risk for dissatisfaction with appearance. The concurrent or proximal factors consist of activating events or situations, which activate schema-driven processing and evaluations. Proximal factors also include body image emotions, and self-regulatory strategies of adjustment. The function of these strategies is to cope with distressing thoughts and emotions that are associated with body image and triggered by environmental events.

In the present study, the investigation of the associations between social experiences and appearance evaluations was given priority. Interpersonal experiences (described as one of the historical influences in Cash’s model), are believed to play a significant role in shaping body image and appearance evaluations. The assumption is that there is a bidirectional link between self-perceptions and other-perceptions. Three processes are considered as important within this interpersonal and sociocultural context: reflected appraisals, feedback on physical appearance, and social comparisons (Tantleff-Dunn & Gokee, 2002). Reflected appraisals refer to other’s opinion of us, or our perceptions of this opinion, and the notion that these appraisals are internalized as our self-evaluations of appearance. Socially reflected appraisal models suggest that a child with a facial difference might internalize other people’s reactions to the disfigurement into his or hers self-views (Thompson & Kent, 2001). Further, it is believed that feedback on physical appearance is one of the interpersonal influences that affect self-evaluations of body image. This association between feedback experiences (such as comments, criticism, staring, or teasing), and appearance-concerns has also been demonstrated empirically (Jones, Vigfúsdottr, & Lee, 2004). In the present context, the question is whether children and adolescents with a facial difference might experience more negative feedback about their appearance, thereby affecting perceptions of self-appraisals, processes that are internalized as negative self-perceptions. Social comparison is yet another influence on self-evaluations of appearance, suggesting that an individual’s appearance comparisons is a predictor of body image evaluations (Lu & Hou, 2009; van den Berg et al., 2007). Hence, whether a child with a visible difference compares him- or herself with media models, with peers, or with other children with a cleft, is expected to have an effect on self-evaluations of appearance.
2.1.2. Visible differences and appearance evaluations

Physical appearance influences social perceptions and interactions, often in accordance with social stereotypes (Cash & Fleming, 2002). In Western societies the stereotype of physical attractiveness linking facial beauty and positive characteristics, or less positive expectations for unattractive others has been widely demonstrated, nonetheless with weaker degrees of effect than expected when explored through meta-analyses (Feingold, 1992; Eagly, Ashmore, Makhijani, & Longo, 1991). Hence, facial attractiveness is a social asset that contributes to social acceptance (Kiyak & Reichmuth, 2002). Beauty seems to be an even more central component of stereotype gender role for the female than the male (Striegel-Moore & Franko, 2002). Empirical evidence has also demonstrated that people behave more favourably towards attractive people than towards less attractive (Langlois et al., 2000). Such stereotypes and sociocultural norms may render individuals with craniofacial anomalies more noticeable and possibly more socially vulnerable (Bull & Rumsey, 1988), as illustrated by laboratory studies of social stereotypes (Shaw, Rees, Dawe, & Charles, 1985). In addition, the increasing use of cosmetic surgery in Western societies (Pruzinsky et al., 2006) may contribute to the existing pressure to be attractive and to strive for a perfect appearance. From a sociocultural perspective, culture defines what constitutes an attractive face (Jackson, 2002). Hence, the degree of satisfaction or dissatisfaction with appearance will depend on the discrepancy between cultural ideals and the person’s perception of him-/herself. However, there are clear individual differences in the degree of vulnerability to sociocultural influences, pointing to the presence of other risk or protective factors.

Faces are always on display, and central in social interaction and communication. According to Cash’s cognitive-behavioural model, physical characteristics are one of the developmental factors influencing self-evaluations of appearance (Cash, 2002). These physical characteristics are clearly affected by the presence of a visible cleft, hence affecting one of the developmental influences that are believed to contribute to an individual’s self-evaluations. Research studies have confirmed that the objective presence of a difference such as a cleft may have a negative impact on judgements of appearance when rated by the affected children (Millard & Richman, 2001; Thomas, Turner, Rumsey, Dowell, & Sandy, 1997; Broder, Smith, & Strauss, 1992), adolescents (Pope & Ward, 1997; Noar, 1991), adults (Marcussen, Paulin, & Ostrup, 2002; Ramstad, Ottem, & Shaw, 1995), and also by independent observers (Feragen, Semb, & Magnussen, 1999; Tobiasen & Hiebert, 1993; Krueeckeberg, Kapp-Simon,
& Ribordy, 1993). Such studies lend support to the theoretical assumptions of internalizations of cultural appearance ideals negatively affecting self-perceptions (Jackson, 2002).

Socially perceived attractiveness is only modestly associated with self-perceptions of attractiveness (Jackson, 2002), pointing to the importance of distinguishing between an objective (physical characteristics) and a subjective appearance factor in the “normal population” (Feingold, 1992) as well as in the research field of visible disfigurement (Moss, 2005). In contrast to lay expectations, it is important to remember that even if the objective presence of a facial difference may be distressful, it is not the type or severity of the facial difference that predicts psychological adjustment (Rumsey, 2002b; Ong et al., 2007). In other words, the understanding and investigation of adjustment to a facial difference involves multifaceted processes, which are often difficult to capture and measure in empirical studies.

2.1.3. Self-perceptions
Since the construct of “self” was described by the classic work of William James (1908), many terms have emerged to describe the fundamental aspects of the self. His contributions have paved the way for future models of a multidimensional and hierarchical self-concept. Further, symbolic interactionists such as C.H. Cooley and G.H. Mead placed their primary emphasis on the social aspects of the self. For Cooley (1902), significant others represent a social mirror (the “looking-glass self”) into which the individual can detect the other’s appraisal of him or her, appraisals that later are internalized as part of the self. Mead (1934) focused on the development of the “generalized other”, being the internalized attitudes and perceived opinions of significant others toward us through social interactions. Both Cooley and Mead believed that the opinions of others shape the self-concept, emphasizing the role of social interactions in self-perceptions.

In the present work, the concept of self-perceptions refers to domain-specific characteristics of the individual (Harter, 1999), in a cognitive-developmental framework, and with focus on the role of social processes in the development of the self. Susan Harter describes the development of early verbal representations of the self, followed by the emergence of self-affects. Social sources of self-evaluations, with the internalization of the opinions of others are further specified. The self is believed to represent a core of inner attributes, consisting of self-evaluative judgements that are differentiated and integrated into a hierarchical model of self-representations. Harter has designed a measure that takes into account these multiple sources of an individual’s view of the self (Harter, 1988), based on a multidimensional
approach of measurement (Marsh & Hattie, 1996). Her scale includes several separate measures of self-perceptions, in domains such as close friendships, social acceptance and physical appearance, which were used in the present work.

Harter (1999) noted that children of 8–11 years come to internalize the standards and values of those who are important to them, in addition to develop an appreciation of sociocultural norms and values. Sociocultural theories stipulates that appearance-related comments from peers about weight or shape (Smolak, 2002), or a child’s differing appearance (Thompson et al., 2001) might affect the child’s self-esteem. This may be explained by the close link between self-perceptions of appearance and global self-worth, an association that is expected theoretically and demonstrated empirically (Harter, 1999; Levine & Smolak, 2002; Lovegrove & Rumsey, 2005; Wichström, 1995). The relation between global self-perceptions and appearance has also been demonstrated in adolescents with a cleft (Bilboul, Pope, & Snyder, 2006). Hence, theory and empirical findings could point to a risk factor of lowered self-concepts when there is a difference in appearance. However, findings are contradictory. Some studies have found lowered self-concepts (Broder & Strauss, 1989; Turner, Thomas, Dowell, Rumsey, & Sandy, 1997; Kapp-Simon, Simon, & Kristovich, 1992; Kapp-Simon, 1986; Leonard, Brust, Abrahams, & Sielaff, 1991), while others have found average or above average self-concept scores (Persson, Aniansson, Becker, & Svensson, 2002; Leonard et al., 1991).

Social functioning and self-evaluations of appearance are undoubtedly intertwined, conceptually, empirically, and experientially (Cash et al., 2002). However, there is a need for more research on protective factors involved in how appearance attitudes affect or are affected by interpersonal experiences when appearance differs from the norms.

2.1.4. Psychosocial experiences and friendships

Social acceptance refers to the degree to which an individual is liked by a peer group, while friendships refer to close, dyadic relationships with specific peers (Nangle, Erdley, Newman, Mason, & Carpenter, 2003). There is a close link between friendships and social acceptance, with overlapping yet unique influences on development and adjustment. Having friends increases the child’s social competence, which in turn increases the social skills that are required to make and keep friends (Hartup & Stevens, 1997). Conversely, research has shown that peer rejection is a risk factor for dysfunctional socialization (Ladd & Troop-Gordon,
2003), possibly prohibiting the development of close, reciprocal friendships. This combination of rejection and lack of positive friendships seems to set the stage for emotional distress (Pedersen, Vitaro, Barker, & Borge, 2007). In contrast, friendship experiences can serve as a buffer between a lack of social acceptance and depression (Nangle et al., 2003).

There is consensus concerning the type of problems that are reported in adjusting to a visible difference, and corresponding difficulties; among the most common problems reported in empirical studies are difficulties with social interaction (Rumsey, 2002a; Clarke, 1999). MacGregor (1979) described a facial disfigurement as a “social disability” since it affects the behaviour of other people in addition to the affected person him- or herself. One of the consequences of a facial visibility would be the inability to feel anonymous and to avoid attracting unwarranted attention.

Research findings point to different aspects of social interaction that might be affected by the presence of a visible difference (Adachi, Kochi, & Yamaguchi, 2003; Frederickson, Chapman, & Hardin-Jones, 2006; Rose & Blakeney, 2006; Kapp-Simon & McGuire, 1997; Richman & Millard, 1997; Slifer et al., 2006). Social psychological research further demonstrated the importance of social skills in the presence of a facial difference, the social skills mediating people’s perception of a disfigurement (Rumsey, Bull, & Gahagen, 1986). Negative experiences such as staring or personal questions may have an emotional impact, in addition to leading them to behave in a less socially skilled manner, such as initiating fewer conversations or making less eye contact (Moss, 1997). Negative social interaction patterns may further contribute to reinforce a negative perception of a person with a facial difference, strengthening a spiralling cycle of problems (Robinson, Rumsey, & Partridge, 1996).

A specific type of stigmatization is appearance-related teasing in childhood (Schwartz & Brownell, 2002). Studies have reported a high frequency of teasing and appearance-related bullying in children with a visible difference such as a cleft lip and palate (Hunt, Burden, Hepper, Stevenson, & Johnston, 2006; Turner et al., 1997; Semb et al., 2005); these experiences might contribute to negative self-appraisals, negative social comparison mechanisms, social isolation, and hence emotional distress. Through experiences of teasing and harassment, children or adolescents may come to the conclusion that they are deficient in attributes that classmates perceive as important and valuable (Ladd et al., 2003). Cross-sectional and longitudinal studies have demonstrated the association between weight-based teasing and emotional distress (Eisenberg, Neumark-Sztainer, & Story, 2003; Eisenberg,
Neumark-Sztainer, Haines, & Wall, 2006; Adams & Bukowski, 2008), an association that has also been found in other visible conditions (Magin, Adams, Heading, Pond, & Smith, 2008). Such findings may prove to be specifically relevant in understanding adjustment to a facial difference such as a cleft, because of the common visibility factor in both conditions (La Greca & Bearman, 2000). Furthermore, studies have reported that experiences of harassment may have a negative effect on perceptions of appearance (Bardone-Cone, Cass, & Ford, 2008; Thompson et al., 2007), an association that has not been demonstrated on individuals with visible differences, as far as it is known.

2.1.5. Emotional adjustment

Research findings indicate that the presence of a visible facial condition may be associated with dissatisfaction with appearance (Millard et al., 2001; Richman, 1997). Negative self-perceptions of appearance carry the risk of emotional distress, a risk factor that is well-known within adolescent samples in general (Holsen, Kraft, & Røysamb, 2001; Paxton, Neumark-Sztainer, Hannan, & Eisenberg, 2006; O'Dea, 2006; Dekker et al., 2007). An association between subjective perceptions of appearance and depressive symptoms has also been demonstrated in samples with other appearance concerns, such as problems related to weight (Eisenberg et al., 2006). However, in spite of the risk of dissatisfaction with appearance due to a facial difference, and in spite of a strong association between perceptions of appearance and depressive symptoms demonstrated in the general population, research findings do not appear to point to a heightened risk for depression in children and adolescents with a cleft. This highlights the need to search for the presence of protective factors, counteracting the negative effect of a potential dissatisfaction with appearance. The question of protective factors has rarely been raised within this field of research, and will be addressed within this work.

While there are no clear indications of heightened risk for depressive symptoms, empirical studies investigating emotional health in individuals with craniofacial anomalies have found generalized anxiety in a significant proportion of adults (Rumsey & Harcourt, 2004; Berk, Cooper, Liu, & Marazita, 2001). Such findings are in line with research from other patient groups affected by a visible difference, such as burn survivors (Rose et al., 2006). However, a study from Denmark indicated a higher incidence of suicide in adults with a cleft than in comparison samples (Christensen, Juel, Herskind, & Murray, 2004). Such findings illustrate that even if emotional health is generally good in children and adolescents with a cleft, some
individuals may be at risk for developing significant emotional distress. This should be kept in mind so that optimal preventive care can be provided.

2.1.6. Developmental patterns
In a developmental framework, risk is closely associated with the challenges inherent in the specific developmental task. The transition from childhood to adolescence is marked by distinct developmental paths, including changes in appearance, changes in friendship dimensions and patterns, and changes related to emotional development. Hence, when investigating adjustment to a visible difference, it is important to keep these developmental patterns in mind.

Research clearly indicates an increase in depressive symptoms during adolescence, primarily among girls (Wichstrøm, 1999; Nolen-Hoeksema, 1994; Graber, 2004). As emotional problems in childhood and preadolescence have been shown to predict later psychiatric disorders (Fergusson, Horwood, & Boden, 2006), it is crucial to identify vulnerability factors that put children born with a cleft at risk for emotional distress.

The most visible and appearance-related developmental change from childhood to adolescence is the onset of puberty, which calls for a reorganization of body image and self-perceptions (Susman & Rogol, 2004). During childhood, global measures of body esteem seem more stable, and there are few differences between boys and girls in their body satisfaction (Smolak, 2002). However, longitudinal studies reveal that girls’ satisfaction with body image declines significantly over the years 12–15 (Levine et al., 2002). This decline in self-perceptions of appearance seems correlated with an increase in emotional problems such as depressive symptoms (Dekker et al., 2007; Paxton, Eisenberg, & Neumark-Sztainer, 2006).

Few studies within the research area of visible differences have large enough samples to study gender differences. One study that did investigate gender differences, did not find the expected decline in satisfaction with appearance in girls (Broder et al., 1992). This could be due to a lower satisfaction with appearance during childhood, compared to their peers. Ongoing treatment possibly also enhances satisfaction with appearance during the adolescent years.

Friendship patterns also change from childhood to adolescence, characterized by an increase of the importance of dyadic interactions from childhood to adolescence (Bukowski, Hoza, &
Boivin, 1993). During the elementary-school-age period, friendships are characterized by less intimacy than what is found during adolescence, and are more often based on common activities, and less on specific characteristics such as self-disclosure and commitment (Newcomb & Bagwell, 1995). In addition, children’s relationships are still characterized by strong parental bonds, and may be more vulnerable to family stressors (Gore & Eckenrode, 1994). It could be that problems related to social acceptance or friendships have a deeper psychological impact during adolescence than during childhood.

2.1.7. Risk and protective factors
Resilience is defined as positive adjustment in spite of risk (Masten, 2001). Risk can be a single event, or more often a sequence of stressful experiences, as in children born with a cleft lip and/or palate. The critical indicators of what constitutes stress must be defined, according to the nature of the risk under study, so that the particular context within which resilience is studied can be established (Luthar, Cicchetti, & Becker, 2000). For a child born with a cleft lip and/or palate, risk is related to one or more of the following associated complications or experiences due to the cleft (Cunningham, 2007; Speltz et al., 2000; Speltz & Richman, 1997; Lockhart, 2003): early feeding difficulties, growth deficiency, chronic ear infections, speech delays or impairments, dental problems, experiences associated with treatment and surgery, and/or the presence of a co-morbid difficulty with possible impact on cognitive and psychomotor development. In addition, risk may be associated with negative psychosocial experiences, or negative self-perceptions as a result of a facial difference and/or divergent speech.

Three main factors are usually mentioned as promoting resilience or vulnerability in the lives of individuals at risk (Luthar et al., 2000): Characteristics of the individual (for example cognitive functioning, personality variables, self-efficacy and self-perceptions), characteristics of the individual’s family (including parenting style and family structure, child-parent relationships, socio-economic status), and the individual’s social context (social relations, friendships, school). Resilience is a product of a mutual influence between these three factors, which may function as potential mediators or moderators of the relationships between stressors, risk and outcome by increasing (vulnerability factors) or decreasing (protective factors) the likelihood of psychopathology (Compas, Connor-Smith, & Jaser, 2004).
The question of resilience in individuals with a cleft was raised early by Clifford (1983), but was not explored until recently. However, during the previous decade, the question of resilience in craniofacial health was raised again, but this has been mainly on a theoretical basis (Strauss, 2001; Eisman, 2001; Mouradian, 2001). A meta-analytic review (Lavigne & Faier-Routman, 1992) of psychological adjustment to pediatric disorders, including cleft lip and palate, confirmed that children with physical disorders were at risk for the development of psychological adjustment problems. The effects of the child’s condition on self-perceptions were less clear. One study, based on a large clinical sample (Pope & Snyder, 2005) points to both risk and protective mechanism in children and adolescents with a craniofacial anomaly.

Empirically-based studies of resilience within this population remain sparse, and mainly based on qualitative methodology (Meyerson, 2001; Edwards et al., 2005; cooper, 2000). However, positive adjustment has been reported in at-risk groups such as burn survivors that also have to cope with the challenge of a visible difference, or survivors of cancer (Lawrence, Rosenberg, & Fauerbach, 2007; Pellard, 2006; Barakat, Alderfer, & Kazak, 2006). An important question in this research area is whether friendships and social acceptance protect against concerns about appearance, which in turn counteract emotional problems. On the contrary, negative social experiences such as peer harassment, might be expected to interact with the presence of a visible difference, and exacerbate concerns about appearance.

2.1.8. Cognitive functioning

An individual’s cognitive functioning is one of the foundations for the development of social competence and psychosocial functioning (Goodman, 2002). A child with a developmental disorder will probably struggle with social skills and interaction difficulties. Consequently, in order to understand the processes that are involved in psychosocial experiences and emotional development, it is necessary to clearly determine the underlying mechanisms affecting the child’s social functioning.

The aim of this study was not the investigation of cognitive functioning in children and adolescents with a cleft. However, this work raises the question of how this central factor might be related to the interpretation of findings within this field of research. Some studies of children with clefts have found a higher than expected proportion of infants at risk for developmental problems, especially among children with cleft palate (Millard et al., 2001; Christensen & Mortensen, 2002; Swanenburg, V, Beemer, Mellenbergh, Wolters, &
Heineman-de Boer, 2003; Speltz et al., 2000; Broder, Richman, & Matheson, 1998; Neiman & Savage, 1997). However, scientific research investigating the neurobiology of cognitive difficulties associated with a cleft has been limited (Nopoulos, Langbehn, Canady, Magnotta, & Richman, 2007), and has seldom explored measures related to psychological and psychosocial functioning. This study maintains that the question of incidence of cognitive difficulties and other associated conditions is closely related to the manner in which research handles the presence of such co-morbid problems (Feragen, Borge, & Rumsey, 2009).

2.1.9. Summary
The importance of distinguishing between objective measures and subjective ratings of appearance has been established. While objective appearance is related to a medical diagnosis, and can be improved by surgical and orthodontic treatment procedures, satisfaction with subjective appearance on the other hand, are psychological in nature. Given the importance of subjective appearance satisfaction for psychological health, a better understanding of corresponding risk and protective factors is warranted. It is also known that various aspects of social interaction might be negatively affected by the presence of a visible difference. However, there have been almost no studies investigating the role of positive social experiences in children or adolescents with a cleft, as a possible protective factor against psychosocial vulnerability or appearance dissatisfaction, a question that is of central importance for clinical interventions and preventive work.

Table 1 provides a brief overview of empirical findings in craniofacial psychological research that have been presented in this section, with information about sample sizes and main findings. Priority was given to relatively recent studies, with only two studies published before 1990. Only studies investigating emotional or psychosocial adjustment, or questions related to appearance have been included.

2.2. The presence of associated conditions
Studies report a varying frequency of associated anomalies in children with a cleft. Such differences can be explained by a discrepancy in the definition of associated anomalies, various types of examination, the child’s age, and how representative the sample is compared to the total cleft population. Empirical studies have reported associated malformations and anomalies in the central nervous system, cardiovascular problems or
syndromes (Milerad, Larson, Hagberg, & Ideberg, 1997; Stoll, Alembik, Dott, & Roth, 2000; Sivertsen et al., 2008). Studies have also indicated an increased risk for learning disabilities in children with a cleft (Richman & Eliason, 1984; Richman, Eliason, & Lindgren, 1988), and an increased incidence of structural brain anomalies (Milerad et al., 1997), with differing degrees of impairment. However, as far as known, no study has investigated the prevalence of associated conditions affecting psychological adjustment in individuals with a cleft lip and palate. The presence of co-morbid difficulties could affect psychological measurements and results if they are not dealt with appropriately, as established in children with physical disabilities (Miyahara & Piek, 2006). The question of how the presence of an associated difficulty impacts on the child’s psychological functioning can only be answered by a careful classification of all associated conditions that co-occur with a cleft lip and/or palate. Hence, it is an important question to address by researchers in this field.

2.2.1. Traditional exclusion criteria in cleft research

There are well-established traditions about dealing with the presence of associated conditions in medical and psychological research. Studies within the field of cleft lip and palate usually exclude those with conditions such as syndromes, mental retardation or those with a significant medical history (e.g. Hunt et al., 2006; Thomas et al., 1997). However, the increased incidence of structural brain anomalies that has been demonstrated (Milerad et al., 1997) indicates that very different degrees of impairment result from such anomalies, and remain undiagnosed. Information about whether a study includes children with ADHD, dyslexia, or other mild cognitive impairments, is usually not provided, even though children with such difficulties are probably included in many published studies. The inclusion of such conditions without considering their presence is likely to affect results and interpretations.
Table 1  Overview of empirical studies investigating psychological functioning in children and adolescents with a cleft (restricted to psychosocial adjustment, emotional adjustment, and appearance evaluations).

<table>
<thead>
<tr>
<th>Reference</th>
<th>Sample size (Cleft + control)</th>
<th>Age</th>
<th>Outcome</th>
<th>Main findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bilboul et al., 2006</td>
<td>49</td>
<td>14–18 yrs</td>
<td>Appearance</td>
<td>Associations between self-perceptions and appearance</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Self-perception</td>
<td></td>
</tr>
<tr>
<td>Broder et al., 1989</td>
<td>58</td>
<td>7 yrs</td>
<td>Self-concept</td>
<td>Children with CLP had lower self-concept scores</td>
</tr>
<tr>
<td>Broder et al., 1992</td>
<td>495</td>
<td>5–18 yrs</td>
<td>Facial appearance</td>
<td>CL/CLP: 54% pleased with appearance and 62% with speech</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Speech</td>
<td>Low but significant correlations between child and parental reports</td>
</tr>
<tr>
<td>Chapman et al., 1998</td>
<td>20 + 20</td>
<td>3–5 yrs</td>
<td>Conversational skills</td>
<td>Preschool children with cleft less assertive conversational style</td>
</tr>
<tr>
<td>Coy et al., 2002</td>
<td>126</td>
<td>3–24 months</td>
<td>Facial appearance</td>
<td>Less attractive infants associated with higher probability of secure attachment</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Attachment</td>
<td></td>
</tr>
<tr>
<td>Edwards et al., 2005</td>
<td>33 adolescents 14 parents</td>
<td>11–18 yrs</td>
<td>Quality of life</td>
<td>Qualitative study: Seven domains identified, such as coping, emotions, self-image, intimacy, and surgery.</td>
</tr>
<tr>
<td>Frederickson et al. 2006</td>
<td>17 + 17</td>
<td>33–44 months</td>
<td>Conversational skills</td>
<td>35% produced fewer assertive utterances and lower responsiveness</td>
</tr>
<tr>
<td>Hunt et al., 2006</td>
<td>160</td>
<td>8–21 yrs</td>
<td>Psychosocial functioning</td>
<td>Participants with CLP reported more psychological and psychosocial problems than controls.</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Teasing</td>
<td>Teasing predicted poorer psychosocial functioning.</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Appearance</td>
<td></td>
</tr>
<tr>
<td>Study</td>
<td>Sample Size</td>
<td>Age Range</td>
<td>Variables</td>
<td>Findings</td>
</tr>
<tr>
<td>-------------------------------</td>
<td>-------------</td>
<td>------------</td>
<td>----------------------------------</td>
<td>--------------------------------------------------------------------------</td>
</tr>
<tr>
<td>Kapp-Simon et al., 1992</td>
<td>45</td>
<td>10-16 yrs</td>
<td>Self-perceptions, Social skills, Social inhibition</td>
<td>Self-perceptions, social skills, and inhibition within normal range</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Adjustment below normal range</td>
</tr>
<tr>
<td>Kapp-Simon et al. 1997</td>
<td>13 + 12</td>
<td>12-14 yrs</td>
<td>Social interaction</td>
<td>Differences in social interaction patterns: Adolescents with CFA more often at periphery of the group, as observers rather than participants in conversations</td>
</tr>
<tr>
<td>Kapp-Simon, 1986</td>
<td>50 + 172</td>
<td>5-9 yrs</td>
<td>Self-concept</td>
<td>Children with clefts lower self-concepts</td>
</tr>
<tr>
<td>King et al., 1993</td>
<td>17 + 36</td>
<td>14-18 yrs</td>
<td>Self-esteem, Self-perceptions</td>
<td>Gender differences: females with physical disabilities lower social acceptance, athletic competence, and romantic appeal</td>
</tr>
<tr>
<td>Krueckenberg et al., 1993</td>
<td>30 + 22</td>
<td>3-6 yrs</td>
<td>Social skills, Attractiveness</td>
<td>Children with CFA rated less attractive and with lower social skills</td>
</tr>
<tr>
<td>Leonard et al., 1991</td>
<td>105</td>
<td>8-18 yrs</td>
<td>Self-concept</td>
<td>98% had average or above average scores, while popularity below norms</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Gender and age effects (adolescent girls' lower self-concept than younger girls. Opposite effect for boys)</td>
</tr>
<tr>
<td>Millard et al., 2001</td>
<td>65</td>
<td>8-17 yrs</td>
<td>Facial appearance and speech, Psychological variables</td>
<td>Children with CP showed greater emotional problems</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Children with CLP showed more appearance-related problems</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Children with UCLP reported lower levels of depression than children with BCLP and CP</td>
</tr>
<tr>
<td>Noar, 1991</td>
<td>32</td>
<td>16-25 yrs</td>
<td>Satisfaction with facial appearance and speech, Teasing</td>
<td>Adolescents satisfied with appearance and speech</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Reports of teasing</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Parents felt adolescents were socially and emotionally affected by the cleft</td>
</tr>
<tr>
<td>Persson et al., 2002</td>
<td>55 + 31</td>
<td>17-20 yrs</td>
<td>Self-concept, Introversion</td>
<td>Adolescents with CLP had normal or higher self-concepts</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td>No introversion</td>
</tr>
<tr>
<td>Study</td>
<td>Participants</td>
<td>Age</td>
<td>Research Focus</td>
<td>Findings</td>
</tr>
<tr>
<td>------------------------------</td>
<td>--------------</td>
<td>-----</td>
<td>----------------------------------------------------</td>
<td>--------------------------------------------------------------------------</td>
</tr>
<tr>
<td>Pope et al., 1997</td>
<td>24</td>
<td>11–14 yrs</td>
<td>Facial appearance, Psychosocial adjustment</td>
<td>Association between social competence, friendships, and self-perceptions of appearance</td>
</tr>
<tr>
<td>Pope et al., 2005</td>
<td>724</td>
<td>2–18 yrs</td>
<td>Behaviour</td>
<td>Behaviour problems, Age and gender patterns</td>
</tr>
<tr>
<td>Richman et al., 1997</td>
<td>44</td>
<td>4–12 yrs</td>
<td>Behaviour, Appearance</td>
<td>Longitudinal study. Increased social inhibition and conduct problems for girls, not related to appearance or speech</td>
</tr>
<tr>
<td>Richman, 1997</td>
<td>65</td>
<td>6, 9, 12 yrs</td>
<td>Appearance and speech, Behavioural problems</td>
<td>At age 9, behavior inhibition and speech problems related. At age 12, inhibition and facial disfigurement related</td>
</tr>
<tr>
<td>Semb et al., 2005</td>
<td>127</td>
<td>17 yrs</td>
<td>Teasing, Satisfaction with treatment, Burden of care</td>
<td>High levels of teasing (74%), Parental reports of teasing (65%), High level of satisfaction with treatment</td>
</tr>
<tr>
<td>Speltz et al., 1993</td>
<td>23</td>
<td>5–7 yrs</td>
<td>Behavioural problems</td>
<td>Minority had behavioral problems</td>
</tr>
<tr>
<td>Thomas et al., 1997</td>
<td>111 patients</td>
<td>10–20 yrs</td>
<td>Facial appearance, Psychosocial adjustment</td>
<td>Patients with visible clefts less satisfied with appearance Appearance and psychosocial adjustment associated Low agreement between patients and parents on adolescents’ perceptions of appearance</td>
</tr>
<tr>
<td></td>
<td>62 parents</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Topolski et al., 2005</td>
<td>56 + 226</td>
<td>11–18 yrs</td>
<td>Quality of life</td>
<td>Adolescents with visible differences lower quality of life than adolescents with other chronic conditions Same for relational domains Higher on family domains than adolescents with mobility limitations or ADHD</td>
</tr>
<tr>
<td>Turner et al., 1997</td>
<td>112 patients</td>
<td>1–20 yrs</td>
<td>Psychological outcomes, Appearance</td>
<td>60% cleft-related teasing Low agreement between patients and parents on perceptions of appearance</td>
</tr>
<tr>
<td></td>
<td>130 parents</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>
2.2.2. Summary

The first consequence of a strong association between a cleft condition and co-morbid difficulties is their potential impact on the measures and variables in a study. This challenge can be dealt with, as proposed throughout this dissertation, by a careful registration of all associated conditions, in order to control for their potential effect on the included measures. Secondly, more knowledge is needed about challenges when the cleft condition is associated with other diagnoses, in order to offer tailored and appropriate treatment and help. Hence, empirical research investigating the psychological impact of these co-morbid difficulties is warranted. However, the obvious heterogeneity in the type and frequency of associated conditions creates the challenge of having large enough samples to study these subgroups.

In view of the lack of understanding of the impact of co-morbid conditions on adjustment, care was taken in this research to document these characteristics in all participants. In all three papers, this question was answered statistically by a thorough registration of all associated conditions, making it possible to control for the presence of a co-morbid difficulty in the analyses. In order to maintain focus on the specific subject within each paper, little information was included about the type of associated difficulties within the samples. Consequently, a more detailed description of the nature and frequency of these associated difficulties is provided when describing the participants in this dissertation.

2.3. Limitations in the research field

Much research on the psychological impact of visible differences is limited by methodological problems such as small sample sizes. Often samples include several different craniofacial conditions (Speltz et al., 1997). Additional methodological problems are related to a lack of longitudinal studies, varying measures and instruments, lack of representative samples, and difficulties in finding appropriate comparison or control groups (Rumsey et al., 2004). Furthermore, a large number of studies are based on samples with widely differing age groups and developmental stages, in order to recruit large enough samples (see Table 1).

Small samples restrict analyses across gender and diagnosis. As a result, in spite of its clinical significance, there is a lack of knowledge concerning differences between boys and girls in their adjustment to a visible difference as a cleft. When interpreting results from studies where such delineations have not been made, it is important to keep in mind this confounding of
gender and diagnosis; visible cleft groups usually include more males, while cleft palate groups have more females (Sivertsen et al., 2008). Results that are interpreted as differences due to a visible difference may thus be a disguised gender difference. The consequence of this population gender difference is that girls with visible clefts and boys with non-visible clefts often will be underrepresented in clinical groups. Very large samples are therefore necessary if these groupings are to yield reliable results.

3. Aims and research questions

The “objective” measure of whether a cleft is visible or not might predict ratings of appearance (Thomas et al., 1997), while its association with psychological adjustment or psychosocial experiences seems more complex. However, as subjective ratings of satisfaction with appearance are known to impact on psychological well-being, the question of the association between objective measures and subjective ratings of appearance is worthy of investigation. There is a need for studies that include both perspectives, with a view to clarify possible interactions between them, and shed light on their contrasting effect.

Research findings support the view that social experiences can affect satisfaction with appearance (Clark & Tiggemann, 2007; Thompson et al., 2007), pointing to processes in which socialization experiences are internalized, affecting the development of self-perceptions (Harter, 1999; Cash, 2002). Assuming that a visible difference affects satisfaction with appearance (Millard et al., 2001) it would be important to ask whether experiences of friendships promote satisfaction with appearance. Most studies have focused on negative experiences, while few studies have tested these associations between positive experiences of friendships, social acceptance, and satisfaction with appearance. Furthermore, there have been no studies that have investigated whether negative social experiences such as peer harassment decrease satisfaction with appearance in children born with a cleft lip and/or palate.

Small samples in craniofacial research have contributed to a lack of knowledge concerning possible gender differences in adjusting to a visible difference. From empirical studies in general psychology, it is widely acknowledged that girls are at risk for appearance dissatisfaction and emotional distress during adolescence (Dekker et al., 2007; Galambos, Leadbeater, & Barker, 2004), a risk factor that could be exacerbated by the presence of a visible difference. On the other hand, girls’ investment in friendships may protect them
against emotional distress and psychological risk (Shih, Eberhart, Hammen, & Brennan, 2006).

An overall goal of this study was to explore possible paths involving objective and subjective perceptions of appearance, and investigate different associations between appearance, emotional well-being, and social experiences. This work investigates whether processes of adjustment and resilience would be comparable to the general population, or point to specific mechanisms that may be related to the presence of a visible difference. Furthermore, this dissertation raises the question of whether the frequency and type of difficulties that are co-variant with a cleft lip and/or palate contribute to the challenges associated with measuring the impact of a cleft on the child’s adjustment.

Hence, this study explores the following research questions:

a) How is the child’s psychological functioning related to type of cleft by comparing children with and without co-morbid difficulties? What factors characterize children classified as psychosocially resilient at age 10 compared to children classified as non-resilient (Paper 1)?

b) Are positive social acceptance and friendships associated with subjective appearance satisfaction in adolescents aged 16? Do subjective ratings of appearance mediate the association between social experiences and emotional distress (Paper 2)?

c) What are the associations between peer harassment and appearance concerns in children and adolescents with a cleft lip and/or palate? How do these associations vary according to the visibility of a cleft, gender, and age (Paper 3)?

4. Methods

4.1. Procedure and research ethics

Individual assessment and psychological counselling are routine within the Oslo team at ages 10 and 16. The age of assessment is determined by recommended multidisciplinary standards of care (surgery, orthodontics, and speech) for children with cleft lip and palate (Shaw et al., 2001). The team’s clinical psychologist conducts the psychological assessments with the child, using a semi-standardized interview. Adolescents fill in a questionnaire after a short interview with the team’s psychologist. Parents also completed a short questionnaire. These
questionnaires (*Questionnaire for 10 or 15-year-old patients* and *Parent Questionnaire*) were developed by the Psychology Special Interest Group of the Craniofacial Society of Great Britain and Ireland, for the purpose of audit and screening. The questionnaires are made up of standardized measures, visual analogue scales, and open-ended questions, derived from relevant theory and research evidence. They are routinely used throughout the UK. A meeting with the parent/s, both before and after the consultation with the child or the adolescent, contributes additional information, if necessary.

Three psychologists were in charge of the assessments that were included in this study, with the assistance of three psychology students. Data from three birth cohorts for each of the two age groups (approximately 450 families) had already been collected when the work on this dissertation began. These families were contacted by post, with a description of the study and a consent form (see Appendix), along with a stamped return-addressed envelope. They were also given the option to replying by email or SMS. Families attending the appointment during this study received the same written description of the study with a consent form, with the option of turning in the form at that time or sending it in later. The psychologist in charge of this study was not involved in this consent form process. Informed written parental consent was collected for children and adolescents below the age of 16. For the adolescents older than 16, written consent was obtained from the adolescents themselves. Parents of adolescents older than 16 were informed and given consent form, in order to determine whether they were positive about their child’s participation.

The Regional Committee for Medical Research Ethics, East, Norway, granted ethical approval for the work of this study. Permission was also granted for the study by the Norwegian Data Inspectorate.

### 4.2. Participants

With a 98–99% turnout for psychological examination, the Oslo team has very few problems with non-attendance. Consequently, the sample of 818 children and adolescents who participated in the 10 and 16-year-old appointments consists of six and five complete birth cohorts, respectively. These birth cohorts include children born 1993–1998, and adolescents born 1988–1992. The sample also includes children born during the second half of 1992.
Due to severe developmental problems, some children and adolescents were not able to participate in the routine evaluations. The only available information for those children was the type of cleft, associated diagnosis, age, ethnicity and gender. The total number of these children was 42 (5% of the children and adolescents within the given birth cohorts). These children and their families were not asked to participate in the study.

All other participants attending the routine appointments were informed about the study and asked to participate. The participation rates were 82% in Papers 1 and 2, and 85.5% in Paper 3. Paper 1 includes child cohorts ranging from 1992–1997, while the child group in Paper 3 consists of children born from 1992–1999. Paper 2 includes adolescents born 1988–1992 while Paper 3 includes adolescents born 1988–1993. Consequently, all participants in Paper 1 (children) and Paper 2 (adolescents) are included in Paper 3 (total sample). Table 2 illustrates the distribution of participants across the three papers.

4.2.1. The comparison group in Paper 2

The clinical adolescent sample from Paper 2 was compared to a large group of same-aged Norwegian adolescents. The comparison group was drawn from the national sample Young in Norway 2002 which comprised 11 371 students from the entire country (Rossow & Bø, 2003), who filled out a self-administered questionnaire at school. The samples originally included adolescents in grades 7–12 from 73 schools. The school samples were stratified according to five geographical regions and according to school size, which is closely related to degree of urbanization. The response rate was 92.3%. All adolescents 16 years of age were drawn from the national sample, making a total of 1832 youths in the comparison group (888 girls, 944 boys).

4.2.2. Prevalence of associated difficulties in the cleft samples

All associated difficulties were recorded as such in the samples. Difficulties were classified as developmental problems, learning difficulties, or “other difficulties”. Developmental problems include diagnoses such as mental retardation or deficit, or problems within the autism spectrum disorder. Learning difficulties include problems such as ADHD, dyslexia, and specific language impairments. The category “other diagnoses” includes hearing impairments or other medical conditions.
Table 2. Distribution of participants across the three papers.

<table>
<thead>
<tr>
<th></th>
<th>Paper 3</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Children</td>
<td>Adolescents</td>
<td>Total sample</td>
<td></td>
</tr>
<tr>
<td>Birth cohorts</td>
<td>425</td>
<td>+</td>
<td>393</td>
<td>→</td>
</tr>
<tr>
<td>Excluded children</td>
<td>18</td>
<td>+</td>
<td>24</td>
<td>→</td>
</tr>
<tr>
<td>Included and informed</td>
<td>407</td>
<td>+</td>
<td>369</td>
<td>→</td>
</tr>
<tr>
<td>Participating sample</td>
<td>346</td>
<td>+</td>
<td>315</td>
<td>→</td>
</tr>
</tbody>
</table>

Type of cleft

<table>
<thead>
<tr>
<th>Type of cleft</th>
<th>Paper 3</th>
<th></th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft lip and palate</td>
<td>198</td>
<td>+</td>
<td>115</td>
<td>→</td>
</tr>
<tr>
<td>Cleft lip/cleft lip alveolus&lt;sup&gt;2&lt;/sup&gt;</td>
<td>23</td>
<td>+</td>
<td>98</td>
<td>→</td>
</tr>
<tr>
<td>Cleft palate</td>
<td>107</td>
<td>+</td>
<td>77</td>
<td>→</td>
</tr>
<tr>
<td>Submucous cleft palate</td>
<td>18</td>
<td>+</td>
<td>25</td>
<td>→</td>
</tr>
</tbody>
</table>

---

<sup>2</sup>Children with CLA, had, until April 2007 not been offered a psychological follow-up at age 10. Thus, only 13 children with CLA were eligible for inclusion in the first paper. They all gave their consent.
Table 3. Frequency of associated difficulties in the child and the adolescent sample of this study.

<table>
<thead>
<tr>
<th></th>
<th>10-years-old</th>
<th>16-years-old</th>
<th>( \chi^2 )</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total</td>
<td>346 (100.0)</td>
<td>315 (100.0)</td>
<td></td>
</tr>
<tr>
<td>No co-morbid</td>
<td>263 (76.0)</td>
<td>268 (85.1)</td>
<td>8.58***</td>
</tr>
<tr>
<td>Developmental</td>
<td>40 (11.6)</td>
<td>16 (5.1)</td>
<td>8.93***</td>
</tr>
<tr>
<td>Learning</td>
<td>30 (8.7)</td>
<td>14 (4.4)</td>
<td>4.74*</td>
</tr>
<tr>
<td>Other difficulties</td>
<td>13 (3.8)</td>
<td>17 (5.4)</td>
<td>1.02</td>
</tr>
<tr>
<td>Syndrome</td>
<td>17 (4.9)</td>
<td>13 (4.1)</td>
<td>.24</td>
</tr>
</tbody>
</table>

Note: Some children or adolescents have a syndrome without any associated difficulties, some have associated difficulties without a syndrome, and some have both.

* \( p<.05 \).  ** \( p<.01 \).  *** \( p<.001 \).

Table 4. Frequency of associated difficulties in children and adolescents with a visible and with a non-visible cleft in the total sample.

<table>
<thead>
<tr>
<th></th>
<th>Visible clefts</th>
<th>Non-visible clefts</th>
<th>( \chi^2 )</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total</td>
<td>434 (100.0)</td>
<td>227 (100.0)</td>
<td></td>
</tr>
<tr>
<td>No co-morbid</td>
<td>371 (85.5)</td>
<td>160 (70.5)</td>
<td>21.22***</td>
</tr>
<tr>
<td>Developmental</td>
<td>29 (6.7)</td>
<td>27 (11.9)</td>
<td>5.22*</td>
</tr>
<tr>
<td>Learning</td>
<td>24 (5.5)</td>
<td>20 (8.8)</td>
<td>2.58</td>
</tr>
<tr>
<td>Other difficulties</td>
<td>10 (2.3)</td>
<td>20 (8.8)</td>
<td>14.56***</td>
</tr>
<tr>
<td>Syndrome</td>
<td>10 (2.3)</td>
<td>20 (8.8)</td>
<td>14.56**</td>
</tr>
</tbody>
</table>

Note: Some children or adolescents have a syndrome without any associated difficulties, some have associated difficulties without a syndrome, and some have both.

* \( p<.05 \).  ** \( p<.01 \).  *** \( p<.001 \).
There was a higher frequency of associated difficulties in the 10-year-olds than in the adolescent sample, as shown in Table 3. Specifically, there was a higher frequency of developmental and learning difficulties in the child group compared to the adolescent group. There were no differences in the frequency of other conditions, such as hearing impairment, or in diagnosed syndromes. Table 4 illustrates the difference in the frequency of associated difficulties between participants with a visible cleft and those with a non-visible cleft. Specifically, there was a higher frequency of associated developmental difficulties, a higher frequency of other conditions, and a higher frequency of syndromes in cases of cleft palate or submucous cleft palate compared to when the cleft was visible.

4.2.3. Ethnicity
In the total sample, 7% of the children and adolescents were of non-Caucasian origin ($n = 47/661$), the majority of them being adopted ($n = 41$, 6.4% of the total sample). In the national comparison sample that was included in Paper 2, 5.3% ($n = 97/1832$) reported that both parents were immigrants.

4.2.4. Attrition
In exploring possible differences between participating and non-participating families in the total sample, it appeared that there was a higher percentage of children and adolescents with a cleft lip and palate (88%) that consented to participate, compared to the children and the adolescents with a cleft palate (80.2%). This difference was statistically significant ($\chi^2 = 8.71, p < .01$). There were also fewer participating families where the child had a syndrome (64.7%) compared to those with no diagnosed syndrome (86.4%, $\chi^2 = 17.63, p < .000$). In the total sample, there were no significant differences between participating and non-participating families with respect to whether there was an associated difficulty in addition to the cleft (81%), or not (86.2%, $\chi^2 = 2.97, p > .05$).
4.3. Measures and instruments

4.3.1. Dependent and independent variables in Paper 1, 2, and 3
In Paper 1, the outcome variable was the child’s perception of social experiences, while the independent variables were objective cleft visibility, gender, presence or absence of a co-morbid condition, experienced teasing and social visibility (questions and staring), subjective appearance satisfaction, emotional functioning, and parental reports of teasing and appearance.

In Paper 2, the outcome variable was depressive symptoms, with subjective perceptions of appearance as a hypothesized mediator of the predictors. The independent variables included the objective measure of cleft visibility, gender, presence or absence of a co-morbid condition, social acceptance, and close friendships.

In Paper 3, the dependent variable was satisfaction with appearance, while the independent variables were objective cleft visibility, gender, the presence or absence of a co-morbid condition, peer harassment, and parental reports of teasing. Frequency of teasing was reported by the child/adolescent and by their parents.

4.3.2. Visibility of cleft
Since the severity and the type of a visible difference does not, in itself, constitute a major marker of psychological adjustment (Thompson et al., 2001; Rumsey et al., 2004; Hunt et al., 2005; Rumsey, 2002b), this knowledge was taken into account when classifying the different types of clefts for the statistical analyses. In other words, whether or not the cleft lip also involved the palate was not expected to affect how the child or the adolescent coped with the challenge of a facial difference. Consequently, types of clefts were categorized as either visible or non-visible in all three papers. Visible clefts include cleft lip and palate, cleft lip alveolus, and cleft lip, whether they are bilateral or unilateral. Non-visible clefts include cleft palate and submucous cleft palate.

4.3.3. Presence or absence of an associated difficulty
As mentioned previously, conditions were categorized as developmental difficulties, learning difficulties, or other problems. For the purpose of the analyses, a dichotomous variable was
created of having or not having a co-morbid difficulty. Consequently, it was possible to perform all analyses while keeping or excluding participants with an additional difficulty within the samples. Analyses could also be performed for subtypes of associated conditions.

4.3.4. Psychosocial experiences, social acceptance, and close friendships

**Social experiences:** In Paper 1, the outcome variable, social experiences, was measured by the *Childhood Experience Questionnaire*, CEQ (Pertschuk & Whitaker, 1982). The CEQ records the child’s self-reporting of social experience on a 5-point Likert scale. The questions in the scale relate to topics such as relations with friends (“I play with friends at school”, “I have friends over to play”), social isolation (“I try to hide from people”, “I keep to myself”), and involvement in new experiences (“I go to new places”, “I meet new people”). Both positively and negatively worded items are included in the instrument, to avoid systematic response bias. Scores are converted to a positive value so that high scores on the CEQ reflect positive social experiences. A mean total score was calculated. The scale has been shown to have satisfactory internal consistency and a coherent factor structure (Emerson, Spencer-Bowdage, & Bates, 2004). In Paper 1, the CEQ showed moderate internal consistency, with a Cronbach’s α value of .68 for the 20 items.

**Teasing, questions, and staring:** Information about teasing, whether the child experienced being questioned about the cleft, and/or experienced staring, was gathered through the patient and parent questionnaires. The questions were answered by yes or no and entered as independent variables in Paper 1.

**Social acceptance and close friendships:** The *Self-Perception Profile for Adolescents*, SPPA (Harter, 1988), is a 35-item scale designed to assess adolescents’ self-perceptions in a number of domains. The Norwegian version was used, which has been shown to achieve better reliability, and better convergent validity than the original version, with a replication of the same factorial pattern (Wichstrøm, 1995). The subscales contain five statements each presented as a self-description. Responses were given on a scale from “Describes me very poorly” (1) to “Describes me very well” (4). A mean score ranging from 1 to 4 was computed.
In Paper 2, the subscales Close Friendship and Social Acceptance were used as independent variables. Examples of the items from the Close Friendship scale include “I have a really close friend I can share things with” and “I find it hard to make friends I can really trust”. Cronbach’s $\alpha = .77$ among adolescents with a cleft, and $\alpha = .75$ in the comparison group. The subscale Social Acceptance contained items such as “I find it hard to make friends” and “Other adolescents find it kind of hard to like me”. Cronbach’s $\alpha = .85$ in the cleft group, and $\alpha = .81$ in the comparison group.

**Peer harassment:** In Paper 3, peer harassment was one of the independent variable. Peer harassment was measured by some statements from the *Child Experience Questionnaire* that was used in Paper 1 (Pertschuk et al., 1982). The structure of the CEQ has been described previously, in the description of the measurement of social experiences. The child’s self-reporting of negative social experience was given on a 5-point Likert scale. Four items reflecting perceptions of social exclusion and peer harassment were identified within the 20 item scale. These statements cover different aspects of negative social experiences such as “I am teased”, “People stare at me”, “I try to hide from people”, and “I’m picked last when others choose sides”. The total score was obtained by summing up the mean score for each item, then dividing this score by the number of statements. The range of the total score was inverted after computing mean scores, so that high scores reflect a high level of peer harassment (Range 0-4). The peer harassment component showed moderate internal consistency in the present study, with a Cronbach’s $\alpha = .63$ (Age 10: $\alpha = .60$; Age 16: $\alpha = .70$).

4.3.5. Satisfaction with appearance

**Satisfaction with appearance:** In Papers 1 and 3, satisfaction with appearance was measured using the *Satisfaction with Appearance Scales* (SWA) which was developed by the Psychology Special Interest Group of the Craniofacial Society of Great Britain and Ireland. The SWA reflects satisfaction with cleft-related and non-cleft-related parts of the face, speech, overall appearance and the visibility of a cleft. Each rating is made on an interval scale of 0 to 10 where a score of 10 indicates very high levels of satisfaction with appearance. A 12-item version of the scale was used in Paper 1, representing one of the independent variables. Cronbach’s $\alpha = .88$. The SWA has been reported to have satisfactory internal
consistency and a coherent factor structure (Emerson et al., 2004). Summing up the scale’s 12 items and then dividing it by the number of items resulted in a mean total score (Range 0-10).

Similar data were obtained from the parents, reflecting their perception of their child’s appearance. These data were entered as one of the independent variables in Paper 1.

Satisfaction with appearance was the dependent variable in Paper 3. Only two of the items from the SWA were administered at both age 10 and 16. These two items measured satisfaction with overall appearance and with the face. Mean scores were computed. Cronbach’s $\alpha = .77$ at age 10 ($n = 346$), and .97 at age 16 ($n = 315$). In the child sample, the mean scores of these two items correlated almost perfectly with the total score including the remaining 10 items that were administered at age 10 ($r = .97, p < .001$). In the adolescent sample, the mean score of the two items also correlated highly ($r = .80, p < .001$) with the subscale Physical Appearance from the Harter *Self-Perception Profile for Adolescents* that was used in Paper 2 (Harter, 1988). Consequently, the use of these two items in Paper 3 was judged as satisfactory for the purpose of this study.

**Self-perceptions of appearance:** In Paper 2, self-perceptions of appearance were measured using a subscale from the SPPA (Harter, 1988). The structure of the SPPA has been described previously, in the description of the measurement of social acceptance and close friendships. The subscale Physical Appearance measures general satisfaction with appearance through five items: “I am not happy with the way I look”, “I wish my body was different”, “I wish my physical appearance was different”, “I think I am good looking”, and “I really like my looks”. Cronbach’s $\alpha = .90$ in the group of adolescents with a cleft and in the comparison group.

4.3.6. Cognitive and psychological functioning

**Cognitive functioning and general adjustment:** Psychological functioning was assessed using the *Personality Inventory for Children*, PIC (Wirt, 1981). The PIC is a multidimensional personality inventory. It provides a good coverage of psychosocial adjustment through various behavioural, cognitive, emotional and interpersonal domains, using the child’s mother as the informant. The PIC provides an empirical classification based on 12 clinical scales, placing a T-value within normal limits, or within the category of mild,
moderate or severe problems. A Norwegian version of the instrument was used (Troland, 1988). There were four subscales used in this study (Paper 1): the intellectual screening scale, the general adjustment scale, and the depression and anxiety scales. The Intellectual Screening scale has been reported to correlate $r = -.55$ with the Full Scale IQ on the Wechsler scales (Wirt, 1981).

4.3.7. Emotional adjustment

*Emotional functioning:* The depression and anxiety scales from the PIC (Wirt, 1981) measured the child’s emotional functioning. These two scales correlated highly in the present work ($r = .81, n = 234, p < .001$), and were thus combined to form one scale. Mean scores were used as one of the independent variables in Paper 1.

*Depressive symptoms:* In Paper 2, the outcome variable, “depressive symptoms”, was measured using 6 items from a shortened version of the *Hopkins Symptom Checklist*, HSCL–25 (Kandell & Davies, 1982). Tambs and Moum (1993) have demonstrated that a strongly abbreviated version of the instrument correlated well ($r = .92$) with the HSCL-25. Kandel and Davies also observed a significant correlation between the 6-item scale and the 90 items of the original symptom checklist (Derogatis, Lipman, Rickels, Uhlenhuth, & Covi, 1974). Each item, such as “Feeling unhappy, sad, or depressed” and “Feeling hopeless about the future”, was rated on a frequency of occurrence during the preceding 14 days, ranging from “Never” (1) to “Very much” (4). Mean scores were calculated. Cronbach’s $\alpha = .84$ in the groups of adolescents with a cleft, and $\alpha = .82$ in the comparison group. Mean values falling below the clinical cut-off score (<1.75) on the HSCL (Rognerud, Strand, & Dalgard, 2002) were interpreted as indicative of emotional resilience in the group of adolescents with a visible difference.

4.4. Statistical analyses

SPSS 16.0 was used in all three papers, while AMOS 7.0 (Arbuckle, 2006) was utilized for model testing in Paper 2. Differences in mean values were tested with t-tests for independent samples in Papers 1 and 3. In cases of two dichotomized variables, differences were tested by chi-square analyses. In Paper 2, differences in means were tested with a Scheffé multiple-
comparison test, due to unequal group sizes. Fisher r-to-z transformations were used to test the significance between correlation coefficients. In Papers 1 and 3, hierarchical multiple regression analysis was used to examine the significance of variables associated with the outcome variables.

4.4.1. Testing of mediation and moderation

Moderation was tested by including the interaction between the two predictors in the last step of the hierarchical regression analysis. To test mediation in Paper 3, the approach of Baron and Kenny (1986) was used. Sobel tests were used to test whether the indirect effects were significant. The Sobel test was conducted by comparing the strength of the indirect effect of the predictor on the outcome variable to the point null hypothesis that it equals zero (Preacher & Hayes, 2004; Baron & Kenny, 1986).

In Paper 2, AMOS was employed for a path analysis, to test the proposed mediational model. The objective of a path analysis is to test whether a theoretical model is supported by sample data. Path analysis has several advantages when compared to more traditional regression analyses in SPSS. One advantage is the possibility of testing alternative models, with corresponding fit indices indicating the degree to which the data fits the model. A second advantage is the possibility to test more complex associations, with multiple predictors in the same regression analysis.

Path analysis was tested using multiple group analyses, in order to test for differences between the two clinical samples and the comparison group. As gender differences were expected in the proposed model, a multiple group analysis was also performed with boys and girls as the two groups. Preliminary analyses were also conducted with multiple group analyses, consisting of subgroups as types of clefts by gender. However, the sample sizes were judged too small to provide reliable results, and were not included in the study. Preliminary analyses were also performed. As empirical findings have pointed to close friendships as a mediator of social acceptance and emotional distress (Nangle et al., 2003), this model was investigated. The data did not fit this model well, strengthening the findings that were presented in Paper 2.

Model-fit criteria that were reported were $\chi^2$, the Normed Fit Index (NFI), the Comparative Fit Index (CFI), the Root Mean Square Error of Approximation (RMSEA), and its lower
(Lo90) and upper (Hi90) ends of a 90% confidence interval. A model was considered to have a close fit if the NFI and CFI were higher than .90, and the RMSEA was lower than .05 (Arbuckle, 2006).

4.4.2. Handling of missing data

The Child Experience Questionnaire was not administered as part of the 10-year-old questionnaire during the first year of psychological assessments for this age group. So, data from this instrument are missing for the first 90 children. These data were missing in Papers 1 and 3. However, the missing data were consequently associated with the first birth cohort upon completion of the semi-structured interview (approximately 90 children). Consequently, there is no reason to believe that these missing data contributed to a bias in results or bias concerning the children included in the sample.

In all three papers, some data were also missing more at random. When filling out the questionnaire, or during the interview, children or adolescents may choose not to answer some items. During the 10-year-old appointment, children fill in the questionnaire during the interview with the team psychologist. In some cases, the child showed tiredness and was not able to continue the length of time necessary to complete the questionnaire. In such cases, some parts of the questionnaire were omitted. The first priority of the semi-structured interview has always been its clinical usefulness and significance. If necessary the process is shortened for the child’s benefit. In most of the cases when the semi-structured interview was too lengthy for the child, it may be that these missing data were not random. It might be that children with psychological problems such as hyperactivity disorders, or mild developmental disorders were more likely to fail to provide full dataset. In Paper 1, there were 41 cases of missing data (total sample = 268), while there were 24 cases of missing data in Paper 2 (total sample = 289). In the comparison group from Paper 2, there were 16 cases of missing data (total sample = 1832). In Paper 3, there were 46 cases of missing data within the child group (total sample = 246), and 80 cases of missing data within the adolescent group (total sample = 315). Hence, in Papers 1 and 2 there were few cases of missing data, indicating that this factor was unlikely to have significantly affected the results of this study. The proportion of missing data was larger in Paper 3, primarily concerning the appearance variable, and mostly in the adolescent group.
In Paper 2, the path model used maximum-likelihood-estimation in handling missing data. This estimation procedure breaks the likelihood function down into components based on patterns of missing data, and thus allows estimation to proceed using all available data (Arbuckle, 1996). As the proportion of missing data was small for both samples, missing data were not expected to affect the results.

5. Results

5.1. Results in Paper 1


The purpose of the first paper was to explore how the child’s psychological functioning was related to type of cleft when comparing children with and without co-morbid difficulties. In addition, this paper investigated the characteristics of children that were classified as psychosocially resilient compared to the children who were classified as non-resilient.

The results indicated that the types of clefts differed significantly with respect to the distributions of subtypes and the frequency of co-morbid difficulties. While 37% of the children with cleft palate had an associated difficulty, this was the case for 22% of the children with cleft lip and palate ($\chi^2 = 7.03, p < .01$). Analyses showed that when excluding all associated conditions, there were no differences in cognitive function or in psychological adjustment between children with cleft palate and those with cleft lip and palate. Psychosocial resilience was associated with adequate emotional functioning ($\beta = -.43, p < .000$), high satisfaction with appearance ($\beta = .29, p < .000$), and a lower frequency of reported teasing ($\beta = -.23, p < .01$). These variables accounted for 32% of the variance in psychosocial experiences. The children that were categorized in the group of non-resilience reported a higher frequency of teasing, questions about the cleft, and of staring. However, only the reported teasing remained significant when all variables were included in the hierarchical regression. Visibility of a cleft, gender, parental reports of teasing or appearance, and co-morbid difficulties did not contribute to an explanation of psychosocial resilience at age 10.
5.2. Results in Paper 2

Adolescents with and without a facial disfigurement: The role of friendships in perceptions of appearance and emotional resilience.

The aim of this study was to test whether positive social acceptance and friendships could strengthen satisfaction with subjective appearance in adolescents with and without a facial difference as a cleft. The study further investigated whether subjective measures of appearance mediated the association between social experiences and emotional distress.

Perceptions of friendships and social acceptance, appearance, and levels of depressive symptoms were compared across the two clinical samples and the comparison sample. The objective measure of disfigurement did not explain levels of emotional distress, while subjective ratings of appearance were predictive. The results revealed that adolescents with a visible difference reported significantly more positive perceptions of friendships and appearance, and a lower occurrence of depressive symptoms than the comparison group. This difference was explained primarily by positive self-perceptions in boys with a visible disfigurement. Girls with a non-visible cleft were more satisfied with their appearance than girls with a visible cleft and girls in the reference group. Subjective ratings of appearance mediated the association between social acceptance and depressive symptoms in all three samples (adolescent with a visible cleft, a non-visible cleft, and the reference group). Model fit indices were excellent, $\chi^2 (3, n = 175, 85$ and 1852) = 3.60, $p > .05$; CFI = 1.000; NFI = .998; RMSEA = .010. Gender did not contribute significantly in explaining the paths between friendships, appearance, and emotional adjustment. Goodness-of-fit measures were adequate, $\chi^2 (3, n = 175, 85$ and 1852) = 8.30, $p = .04$; CFI = .997; NFI = .996; RMSEA = .029. In the last model, paths were constrained to be equal, in order to investigate whether the three groups differed in the way appearance mediated the associations between social acceptance and depressive symptoms. The goodness-of-fit indices indicated that the model could be applied on the three groups, $\chi^2 (10, n = 175, 85$ and 1852) = 18.86, $p < .04$; CFI = .991; NFI = .996; RMSEA = .021.
5.3. Results in Paper 3
The role of peer harassment when appearance differs

The purpose of the third paper was to explore the associations between peer harassment and appearance concerns in children and adolescents with a cleft lip and/or palate. The study further investigated how these associations varied according to the visibility of the cleft, gender, and age.

While 25% of the children with a non-visible cleft reported teasing, 41% of those with a visible cleft reported such negative experiences ($\chi^2 = 6.59, p < .02$). Among the adolescents that reported previous teasing, there were no differences related to the visibility of the cleft ($\chi^2 = 0.44, p > .05$). The parents reported a lower frequency of teasing of their child than the children or the adolescents themselves ($r = .26, p < .001$). Age and gender differences were found in ratings of appearance, with adolescent girls reporting lower satisfaction with appearance, $t (88) = 2.27, p < .03$, and higher levels of harassment, $t (120) = -2.83, p < .01$, than girls with a non-visible cleft. An interaction between gender and cleft visibility was found ($\beta = .28, p < .05$), affecting ratings of appearance in a negative way in girls with a visible difference. Gender differences in the adolescent sample were demonstrated, with girls being less satisfied with appearance, $t (234) = -3.41, p < .001$, and reporting more harassment than the boys, $t (310) = 3.28, p < .001$. Cleft visibility did not differentiate adolescent boys, or children aged 10 on the study variables. Finally, there were clear associations between harassment and appearance-concerns in all groups, except girls aged 10. Parental reports of teasing did not explain any variance in satisfaction with appearance. Experiences of harassment explained 6% of the variance in appearance scores at age 10, while explaining 23% of the variance in appearance scores in the adolescent group. Experiences of peer harassment totally mediated ($z = 2.39, p < .02$) the association between the visibility of a cleft and appearance ratings in the adolescent girls, $F (1, 89) = 12.25, R^2 = .22, p < .001$.

6. Discussion
The primary goal of this study was to examine possible paths involving objective and subjective perceptions of appearance, while exploring different associations between appearance, emotional adjustment, and social experiences in children and adolescents with a cleft. Further, this dissertation raises the question of whether the frequency and type of
difficulties that co-varies with a cleft lip and/or palate contributes to difficulties in measuring the impact of a cleft on the child’s adjustment.

Results from all three papers indicated that the objective measure of cleft visibility did not differentiate between children and adolescents that coped well compared to those who did not, while the subjective ratings of appearance did.

Results further indicated that adolescent boys with a visible difference reported significantly more positive perceptions of friendships and social acceptance, and a lower occurrence of depressive symptoms than same-aged boys from the comparison group. This finding was interpreted as an indication of emotional resilience. In contrast, adolescent girls with a visible cleft were at risk for appearance dissatisfaction and increased levels of peer harassment in comparison to adolescent girls with a non-visible cleft, and to boys of the same age. However, when comparing adolescent girls with a visible cleft with same-aged girls without a cleft, there were no indications of psychological vulnerability. Hence, the question is whether the group of girls with a non-visible cleft stand out as different in any specific way.

Further, results demonstrate that gender differences are associated with age, indicating a developmental shift in vulnerability and resistance. There were no differences between boys and girls in children aged 10, while gender differences were found in adolescents. In line with theoretical expectations and empirical findings, adolescent girls reported more depressive symptoms, lower satisfaction with appearance, and higher levels of harassment than boys. Another gender difference was that peer harassment mediated the association between the visibility of a cleft and appearance ratings only in adolescent girls. There were no differences, however, between boys and girls on the path between social acceptance and emotional adjustment that was mediated by appearance.

Types of clefts differed significantly with respect to the frequency of co-morbid difficulties. The presence of an associated condition clearly affected cognitive function, general adjustment, and emotional functioning, when information was provided by the child’s mother. However, the presence of a co-morbid difficulty did not affect the outcome measures in the papers.
6.1. Methodological issues

6.1.1. Measurement issues

The measures used in this study were restricted by the instruments in use during the routine psychological assessments at ages 10 and 16. In Paper 2, the measures chosen were those that were included both in the routine psychological examination of adolescents with a cleft and the national survey from 2002. Whether or not there is a facial difference, adjustment involves a complex combination of other psychological and social factors that were not included in this study. Variables that are known to be important include individual characteristics and personality (such as self-efficacy and temperament), cognitive processes (such as attribution style, importance of appearance schemas, internalized social expectations), family factors and social support, as well as socio-cultural factors (Luthar, 2006; Strauss & Fenson, 2005).

During adolescence, it is also known that involvement in a romantic relationship constitutes a developmental change that affects adjustment and friendship experiences (Brendgen & Vitaro, 2002; Kuttler & La Greca, 2004). Such variables should be investigated in future research concerning the area of visible differences.

In addition, the issue of measurement is related to the data source. In this study, data at age 10 was based on a semi-structured interview, while a questionnaire format was used at age 16. Both methods may have influenced the information that was obtained. Interviews are sensitive to interviewer bias, while questionnaires may facilitate honesty or avoidance in the participant (Cox, 1995).

Validity characterizes the extent to which an instrument is capable of measuring what it is supposed to measure, while reliability refers to the consistency of a measurement. Psychometric properties of the specific measures were given in the Methods. However, an overall judgement of whether the chosen measures were adequate in this study is also needed. Internal consistency for all scales was identical or somewhat higher in this study than reported in Wichstrøm (1995). The factorial validity of the Norwegian version of the SPPA (Harter, 1988) was tested by Wichstrøm (1995), and confirmed in the revised Norwegian version. The exception was that the physical appearance scale and the global self-worth scale could not be separated. This merger of the two scales is acknowledged (Aasland & Diseth, 1999; Harter, 1999), while it is not clear whether this is explained by a causal relationship between appearance and global self-worth, or whether the scales measure the same domain. Because only the appearance scale was used in this study, this problem was judged as not relevant in
the interpretation of results. Convergent and discriminant validity were also shown to be adequate for all subscales (Wichström, 1995).

The use of multiple regression to estimate mediation requires that there is no measurement error in the mediator (Baron et al., 1986). This assumption was perfectly met for the testing of appearance as a mediator. However, the measurement of peer harassment showed moderate internal consistency in this study, especially in the child group. The paths involving the mediator may hence be biased by reduced reliability, with an overestimation of the mediational path as a consequence. However, in this study, mediation was only tested in the adolescent sample of girls. Internal consistency was therefore tested within this group, and showed acceptable reliability ($\alpha = .78$) for the purpose of this analysis.

Self-reports are by some researchers considered as less valid than more “objective” reports (Rønning, Handegaard, & Sourander, 2004). Self-reports may be vulnerable to response style or to self-presentation biases, an effect that has been demonstrated in children with chronic illness (Phipps & Steele, 2002). Self-reports may also be vulnerable to shared method variance. Hence, it is recommended that variables are assessed from multiple sources (Hawker & Boulton, 2000). For example, studies on friendships have indicated that there is a need to specify the reciprocity in friendships if we are to understand their impact on psychological development (Bukowski, Brendgen, & Vitaro, 2007), a question that is important also in the present work. On the other hand, the results of the present study indicate how crucial the individuals’ own perceptions of appearance are in the search to understand the impact of a visible difference on adjustment, a perspective that was confirmed by the fact that parental measures did not predict psychosocial adjustment or the child’s satisfaction with appearance. In addition, clinical experience indicates that some children and adolescents with a cleft find it difficult to talk to their parents about their inner feelings of difference, maybe in order to avoid parental distress (Bradbury, 2002). Hence, self-reports, as used in this study, may be among the reliable sources of information about internal emotional states (Ladd et al., 2003).

Another measurement issue that Harter recommends, is additional consideration when investigating concerns self-perceptions in special populations (1999, p.137ff.). Not enough is known about those whom adolescents with a visible or a non-visible cleft choose to compare themselves with when answering the statements in this questionnaire. Their choice of comparison may affect their scores differentially compared to individuals from the general
population. It could be that the unexpected differences between adolescents with a non-visible cleft and the two other groups illustrate the recommendation given by Harter.

Socio-economic status, divorce, and parent education level should have been included in this study, in light of their known impact on psychological adjustment (Wallander, Varni, Babani, Banis, & Wilcox, 1989). The parent’s occupation is noted in the child’s case records soon after birth. However, it is rarely updated, and was thus considered to be not representative of the actual level of education. Their potential impact on the results of this study was considered to be low, given that socio-economic status and educational level are expected to have a reduced impact in Norwegian samples than in many other Western societies (Heiervang, Goodman, & Goodman, 2008). In addition, a meta-analysis (Lavigne & Faiers-Routman, 1993) did not identify socio-economic status as a correlate of significant stressors in children’s adjustment to pediatric physical disorders.

A final measurement issue is whether the measure of social acceptance and close friendships actually was a valid measure of social experiences or not. The goal of this study was to search for variables that could help us predict psychological functioning in children and adolescents with a cleft. A recent study demonstrated that a positive internal sense of social acceptance as measured by the Harter scales (subjective measure) was equally important in predicting social functioning as the more objective ratings of popularity (McElhaney, Antonishak, & Allen, 2008). Such results hence support the notion of measurement validity of the chosen measure when assessing the child’s or the adolescent’s subjective perception of well-being.

6.1.2. Lack of control group

While the results and interpretations were strengthened by the presence of a reference group at age 16, there was a lack of a comparison group for the children. Because of a centralized treatment setting, children that are assessed in Oslo represent regions from the whole country, except those served by the team in Bergen. Consequently, in order to find a representative national reference group, it would have been necessary to stratify participants according to level of urbanization and geographical regions. This was not achievable during the work on this study. Care should be taken in the future to include instruments and measures that are in use in comparable research studies, allowing comparisons between national samples.
The issue of lack of child control group was solved by using within-group analyses. The children and the adolescents with a cleft palate were used as controls for those with a visible difference of the face. The question is whether this is an acceptable choice. One study stated that individuals with cleft palate might have subtle facial anomalies that affect attractiveness ratings in a negative way (Coy, Speltz, & Jones, 2002). However, Coy et al. (2002) excluded only children with diagnosed syndromes, and not those with other associated conditions. As has been demonstrated in the present work, many other conditions were probably included in the study. The present work maintains that when associated conditions or difficulties are carefully identified and registered, children with a cleft palate may be used as controls for a visible condition.

6.1.3. The issue of causality
All three papers made theoretical and statistical assumptions of causality, despite dealing with cross-sectional data. In each paper, a model of causality was proposed, and tested, a model that was based on theory and previous empirical findings. However, the results are not presented as one-way truths. The directionality of the models was chosen in order to shed light on processes believed to be particularly important at the given developmental stage. Hence, friendships and social experiences were expected to predict the risk for depressive symptoms in adolescence (Oldenburg & Kerns, 1997), while depression and anxiety in the child was treated as one of the predictors at age 10. Depressive symptoms as measured in the child group were believed to reflect more stable temperamental characteristics within the child because of the chosen instrument (Wirt, 1981), an inventory that is empirically constructed. In contrast, the adolescents were asked to report symptoms occurring during the last two weeks (Kandell et al., 1982). Consequently, it is assumed that the measure could be more reactive to recent social experiences than the personality inventory, and hence it could be assumed to measure the outcome of the adolescent’s perceptions of friendship and social acceptance.

Longitudinal research will be necessary to confirm the directionality of the relations between the variables reported in the three papers. Depressive symptoms may obviously interfere with the formation of friendships and self-perceptions of appearance (Zimmer-Gembeck, Hunter, Waters, & Pronk, 2009), and intensify feelings of loneliness, thus unfavourably impacting friendship quality and social acceptance. On the other hand, difficulties in peer relationships
may exacerbate emotional difficulties (Pelkonen, Marttunen, & Aro, 2003; Galambos et al., 2004). Furthermore, emotional distress has been shown to facilitate memories of negative social experiences such as teasing (Roth, Coles, & Heimberg, 2002), underlining the importance of longitudinal studies. However, in all papers, longitudinal studies were reported that strengthened the causal interpretation of the results.

6.1.4. Representativeness and generalizability of the sample
A review article (Hunt et al., 2005) systematically searched for articles on the psychosocial impact of a cleft lip and palate on children and adults. Among the articles reviewed, half of these had less than 80 participants, while only five articles had more than 200 participants. It is well-known that small samples limit statistical power, and prevent analyses of outcome by subgroups such as gender or specific diagnosis. Small samples also restrict interpretations and generalization of findings. Hence, one of the strong points of this study is its large and representative sample due to a centralized treatment setting, and almost no problems with non-attendance. The national reference group in Paper 2 was also large and representative, in addition to providing data from same aged-adolescents. The inclusion of children with co-morbid diagnoses while controlling for its potential effect on the outcome variables is also considered to be a strong point in this study. As a result, the sample is more representative of the whole cleft population than in most studies within this field of research.

Attrition was higher in the group of children with cleft palate (20% non-consenting) in comparison to those with a cleft lip and palate (12%). The fact that 88% of the children and adolescents with a visible cleft from 5–6 consecutive birth cohorts for each age group contribute to a unique clinical sample, strengthening the findings that are directly related to this group. A higher attrition rate in the cleft palate group is probably explained by the fact that parents and adolescents believed the study was aimed primarily at children with a visible cleft. Attrition analyses also revealed that there were less families consenting to participate when the child was diagnosed with a syndrome. These differences indicate that children with a cleft palate and children with a diagnosed syndrome are the groups that may be less representative of the population under study. Hence, it might be that attrition affected the results. The unexpected findings related to girls with a non-visible cleft in Paper 2 may partially be explained by attrition within this group.
The question of whether findings from this study can be generalized to other visible conditions is a matter of discussion. It seems that there are reasons to choose a generic approach across conditions rather than a condition-specific approach (Lavigne et al., 1992; Kish & Lansdown, 2000), indicating that findings such as those presented in here can be generalized to other conditions affecting facial features, such as congenital conditions (such as syndromes and birth-marks), disease (such as cancer and vitiligo), or conditions acquired later in life (such as burns or accidents).

Whether the results from this study can be generalized to cleft populations in other countries is a matter for debate due to general challenges associated with comparisons across nations (Alsaker, Flanagan, & Csapó, 1999). First, the organization of cleft care differs from one country to another (Shaw et al., 2001). The importance of long-term appointment by multidisciplinary teams, as practiced in Norway, is recognized as significant in conditions such as major burns or craniofacial anomalies (Sheridan et al., 2000; Kish et al., 2000; Lockhart, 2003). In addition, centralized treatment settings strengthen the external validity of the results (Speltz, Kapp-Simon, Cunningham, Marsh, & Dawson, 2004). In addition, the Norwegian welfare system provides full financial support for all aspects of cleft treatment and treatment-related expenses. This is in contrast to treatment settings where there are no multidisciplinary and specialized teams, and where treatment choices are not based on experience and research (Shaw et al., 2001). Other differences may be related to an association between frequency of cleft conditions and social disadvantage, as has been reported in the UK (Speltz et al., 2004). An epidemiologic study investigating the influence of socio-economic status on the frequency of birth defects in general concluded that paternal educational level was associated only with a slightly increased risk (Stoltenberg, Magnus, Lie, Daltveit, & Irgens, 1997), pointing to some possible differences between Norwegian and British cleft samples. When comparing measurements of psychological functioning across borders, such differences should be addressed and interpreted.

Ethnically speaking, the Norwegian population is relatively homogeneous, consisting of a large majority of Caucasian children and adolescents, as illustrated through both the clinical sample and the comparison group. Consequently, the results from this study may not apply necessarily to other ethnic groups and cultural settings, with possible differing attitudes or beliefs about the meaning and consequences of a visible difference. As far as known, there have been no studies investigating potential differences in attitudes toward visible differences or psychological adjustment between various ethnic groups. Recent research indicate that
there is a movement toward less ethnic differences in weight-related problems compared to some decades ago (Roberts, Cash, Feingold, & Johnson, 2006), a result that may apply also to the potential impact of a facial difference across cultures.

6.2. The presence of associated difficulties

Results from this study indicate that there is a difference in the frequency of reported associated difficulties in the two age groups. This could be explained by the differences in setting for the psychological assessment: While the interview with the child and the contact with the parents facilitated access to information concerning the possible existence of associated difficulties, the psychologist met the adolescents and their parents more briefly. The variations in the frequency of associated conditions may hence reflect how different treatment settings and differences in types of examinations affect the degree to which mild cognitive impairments or other more subtle difficulties are identified or go unnoticed. Whether such conditions are identified will clearly depend on method of assessment, and whether or not psychological assessments are part of routine examinations.

Another interpretation could be that some adolescents who were identified with developmental delay at age 10 have caught up with their peers by age 16. The fact that there were no differences in the frequency of other diagnoses or syndromes may strengthen this hypothesis. However, the lack of difference between these specific conditions may also be explained by the fact that they are easier to discover, and consequently more accurately diagnosed regardless of the assessment setting.

Regardless of the reason for the difference in the frequency of associated difficulties between the two age groups, the results confirm the need of a careful description and registration of related medical or psychological conditions (Cunningham, 2007), in order to distinguish between the consequences of a cleft and the consequences due to an associated difficulty. Classifications have to be done according to clearly defined criteria, which in turn facilitate comparisons between studies. Result clearly showed that the presence of an additional difficulty significantly differentiated between children with cognitive difficulties, general adjustment problems, and emotional difficulties when rated by the child’s mother, while cleft type did not. Preliminary statistics that were not included in the papers also indicated that the presence of an additional difficulty affected subgroups within the samples (such as girls with cleft lip alveolus) on some of the study variables. However, due to the confounding of type of
cleft and gender, these subgroups were judged as too small to provide reliable results. This issue, which is clinically significant and important, should be addressed in future research, in order to effectively identify possible risk groups, and be able to develop appropriate treatment and assistance.

It could be that some problems in psychological adjustment reported in previous research are actually an effect of a negative bias partly explained by the presence of undiagnosed, mild additional difficulties in the samples. As shown in this study, when excluding all identified additional conditions, there were no differences in general adjustment or cognitive functioning between children with a cleft lip and palate and children with a cleft palate. If exclusion criteria had been limited to children with diagnosed syndromes or mental disorder, as in the majority of reported studies, differences between the two types of clefts in measures of adjustment would have been reported in the first paper. This may be explained by the higher frequency of problems associated with cleft palate, masking coping and positive adjustment in children with a cleft when there is no additional difficulty. However, the choice of the outcome variable, or the data source, can also be expected to affect the results. The presence of an associated difficulty affected the outcome measures when rated by the child’s mother, while the measures based on self-reports did not differentiate between children with and without additional problems. This study hence stipulates that the starting point for all research related to craniofacial anomalies should be a thorough registration of associated difficulties and diagnoses, making it possible to control for this factor, and gain a better understanding of the consequences of such associated difficulties.

6.3. Adolescents with a visible versus a non-visible cleft

Adolescent girls with a non-visible cleft reported significantly more positive self-perceptions of appearance than the comparison group. This was an unexpected finding, with various possible interpretations. Adolescents with and without a visible cleft completed the questionnaire in the same room, a factor that might have affected their ratings. As stated by Harter (Harter, 1999), there is not enough known about whom the adolescents from the clinical sample compared themselves with. Adolescents with a visible difference and the comparison group might both have reported their satisfaction with general appearance, such as body mass and shape, while the adolescents with a cleft palate might have been prompted by the presence of the adolescents with a visible cleft in the same room to interpret the
statements as being related to facial features. However, this difference was not found in the group of boys. Hence, the results may indicate a gender difference in adolescents with a cleft palate, or it may be an effect of measurement error due to the small group of boys with a cleft palate. This finding demonstrates the importance of selecting appropriate measures of assessments, and to specify the aspects and the dimensions of body image which are assessed.

Adolescents with a non-visible cleft also reported more negative perceptions of social acceptance than adolescents with a visible cleft and the comparison group. The lower frequency of developmental difficulties in the adolescent sample compared to the child sample could indicate that subtle impairments and difficulties were not identified in the adolescent sample. The presence of such impairments would have the potential to impact on perceptions of social acceptance. Clinical experience indicates that the parents and adolescents focus on the lack of visibility of the cleft. Consequently, subtle impairments may constitute a risk factor without being recognized as such by the individuals themselves or their families, while still affecting adjustment and self-perceptions. On the other hand, analyses controlling for the presence of the co-morbid difficulties that were recorded did not affect the study variables.

Finally, the difference in the group of adolescents with a cleft palate could be due to the confounding effect of speech difficulties as a result of the cleft palate. Speech difficulties could be expected to affect social experiences and emotional adjustment. On the other hand, recordings and clinical experience indicate a very low frequency of speech difficulties at age 16 in Norwegian cleft samples (Kjøll, Tordal, & Flaa, 2005).

6.4. Psychosocial experiences, friendships and social acceptance

Studies investigating appearance-related distress in normative populations of adolescents in the UK have illustrated that concern related to appearance are widely present, and may not be specific to individuals with a visible difference (Lovegrove et al., 2005). Still, as many as 60–75% of children with a visible cleft reported appearance-related teasing in previous studies (Turner et al., 1997; Semb et al., 2005), a disturbingly high percentage. Results from the present study indicate less teasing and bullying than what was reported by Turner et al. (1997) and Semb et al. (2005), and less than what was reported from a large regional sample
of Norwegian children (Rønning et al., 2004). However, it should be noted that the frequency of reported teasing among children with a visible cleft in this study was higher than when the cleft was not visible. The facial visibility may thus have triggered negative social experiences.

Norway has a strong tradition of combating bullying and teasing in schools (Tikkanen & Junge, 2004). Thus, one interpretation could be that there is a higher acceptance for a facial difference in Scandinavian countries compared to Britain. However, the high frequency of teasing that was reported by Semb et al. (2005) also included results from Scandinavian countries, including Norway. Another explanation is that differences in frequency of teasing is related to how teasing is defined, measured, and interpreted.

As in most studies comparing child and parental reports (Borge et al., 2001), parents indicated lower levels of teasing than the child. Children and parents might actually differ in their interpretation of teasing. Another interpretation is that parents are not always aware of ongoing or previous teasing, or they may underestimate the impact of peer comments. Importantly, comments may be interpreted differently by the child him- herself, hence be experienced as teasing by vulnerable children while being interpreted as a neutral question by a secure child. Regardless of the chosen interpretation, the findings emphasize the importance of carefully addressing the child’s opinion and perception of psychosocial experiences independently, in addition to those of their parents.

Higher frequency of harassment in adolescent girls could indicate a vulnerability factor in the presence of a visible difference, due to the associations between victimization, depression, and body dissatisfaction (Paxton et al., 2006; Eisenberg et al., 2006; Hawker et al., 2000). However, findings from this study showed that girls with a visible cleft were not more at risk for depressive symptoms than girls from the comparison group. Viewed together, these findings suggest the presence of protective factors. In spite of the psychosocial risk related to harassment in the presence of a facial difference, positive perceptions of friendships and social acceptance may concurrently strengthen appearance satisfaction, and protect against emotional distress. This is in line with studies demonstrating the protective effect of close friendships in preventing the negative consequences of harassment (Hodges, Boivin, Vitaro, & Bukowski, 1999), a buffering effect that was not found in another study (La Greca & Harrison, 2005). These contrasting findings may shed light on the influential effect of peers on predicting body image as related to the presence of vulnerability or resistance factors within the child. A study demonstrated that the effect of peer influence was present only for
overweight and at risk overweight girls (Thompson et al., 2007). Differences in findings between studies may also be explained by a differing impact of harassment on the child. A longitudinal study demonstrated that harassment impacted on emotional well-being first of all if early experiences of teasing had affected the child’s emotional well-being, and whether there was a history of further teasing (Eisenberg et al., 2006; Kochenderfer-Ladd & Skinner, 2002). Such results point to the importance of differentiating between frequency of teasing, and the emotional impact of those experiences (Kvalem, von Soest, Roald, & Skolleborg, 2006).

According to developmental differences in the importance of friendships, the significance of social acceptance decreases from late childhood, while friendships become more central (Gifford-Smith & Brownell, 2003; Bukowski et al., 1993). The results suggest that the feelings of intimacy and closeness provided by friendships are strongly connected to perceptions of popularity and social acceptance. Hence, the association that was found between social acceptance and lack of depressive symptoms was expected also for perceptions of close friendships. A possible interpretation could be related to the measure of close friendships, tapping the adolescents’ perceptions of this variable, and not necessarily the objective presence of friendships. Measures capturing the reciprocal dimension of social relations should be included in future research, in order to assess various aspects of friendships.

It could also be that these findings are a result of the chosen outcome variable. The nature of appearance-concerns in the presence of a facial difference has been shown to be associated with measures of social anxiety (Rumsey, Clarke, White, Wyn-Williams, & Garlick, 2004), while few studies have reported a risk for depressive symptoms in this patient group (Rumsey et al., 2004). Given that experiences of harassment are social in nature, one might expect it to be related to social emotional problems, such as anxiety. Consequently, subsequent studies should include such measures when investigating emotional adjustment. On the other hand, a review paper reports no evidence supporting the assumption of a link between victimization and anxiety, while supporting an association between victimization and depressive symptoms (Hawker et al., 2000), strengthening the choice of the outcome measure in the present study.

The role of social acceptance on emotional adjustment in adolescents was explored without including information about teasing or peer harassment in the same paper. Since half of the adolescents reported previous harassment, it would have been interesting to investigate the
proposed mediational model of appearance according to previous teasing experiences. Children classified as psychosocially resilient reported less teasing, indicating that the paths from social experiences to appearance might be stronger for adolescents experiencing appearance-related teasing and harassment. This question should be addressed by future research.

6.5. Social cognitions and attributions

Clinical experience and empirical findings confirm the social visibility aspect of facial differences, while also demonstrating great individual variability in adjustment to this difference (Lansdown, Rumsey, Bradbury, Carr, & Partridge, 1997). The child’s ability to experience questions from peers as non-threatening could be one factor at work, minimizing the effect of potential negative social experiences. Such interpretations would be in line with the concept of self-schemata, as proposed by Hazel Markus (1977). Self-schemata are described as cognitive generalizations about the self, derived from past interpersonal experiences. Hence, children who have developed positive core self-evaluations may interpret interpersonal experiences and sociocultural expectations as less threatening, compared to children that experience their facial visibility as central to their appearance-related evaluations. This interpretation is also in line with Cash’s cognitive-behavioural model (Cash, 2002).

Social cognitions and children’s social processing biases (attributions) have been shown to explain differences in how individuals cope with risk (Crick & Dodge, 1994), and demonstrated associations between (experiences of -) teasing, attributional style and appearance (Thompson et al., 2007). Crick and Dodge propose that children are presumed to process external stimuli through a series of steps, including encoding and interpretation of cues. “Objective” experiences are filtered through the individual’s brain via perceptual and cognitive processes (Deater-Deckard, Ivy, & Smith, 2006). Empirical evidence has also demonstrated that negative emotions trigger the memory of negative events (Joormann, Teachman, & Gotlib, 2009), demonstrating the importance of including background information about emotional adjustment. Attributional processes and social cognitions may thus explain some of the individual differences in the manner children and adolescents with a visible difference cope and experience social visibility. It could be that attributional style mediates the link between other peoples’ reactions to the individual’s difference, and the
effect these reactions may have on their emotional and psychosocial adjustment. The data from this study does not lend itself to test for this assumption. However, since self-reports may be vulnerable to attributional style, this discussion seems warranted. Future research should address the questions of attributional style in coping with a visible difference.

How subjective body image may affect our emotional and cognitive processing of social interactions was elegantly demonstrated by Kleck and Strenta (1980). Participants that thought they had a facial scar made up by theatrical makeup reported more negative social reactions in an interaction setting than participants that did not believe they had been disfigured by the way of makeup. For children with a visible difference, comments, questions and gazes may hence be interpreted as negative, neutral or positive interest, interpretations that are likely to be related to the child’s self-concept and esteem.

6.6. Gender differences

Results from this study shed light on possible developmental differences in the way boys and girls may adjust differently to a visible difference of the face. Differences between adolescent boys and girls are consistent with theory and previous empirical findings, indicating a heightened risk for depressive symptoms, dissatisfaction with appearance, and higher levels of peer harassment (Dekker et al., 2007; Galambos, 2004; Leenaars, Dane, & Marini, 2008).

The assumption that gender could have the potential to moderate the effect of a facial difference on satisfaction with appearance was also demonstrated in the present work, an effect that was not found among adolescent boys. This finding seems to be opposite to what was reported by Leenaars et al. (2008), a study that demonstrated that attractiveness could appear to function as a protective factor against victimization for boys, while being a risk factor for adolescent girls. According to Leenaars et al. (2008), one could ask whether the presence of a visible difference such as a cleft hence might constitute a protective factor for girls, while the opposite effect would be the case for boys. However, attractiveness and satisfaction with appearance are not highly correlated (Jackson, 2002), impeding a comparison of Leenaars’ findings and this study.

The importance of interpersonal experiences both as a protective and as a risk factor for self-evaluations of appearance was demonstrated in this study, in line with Cash’s cognitive-behavioural model of body image (Cash, 2002). Gender differences that were found in this
study may point to differential influences of historical factors, and differential attitudinal dimensions of self-evaluations. The interaction effect between gender and visibility of cleft in girls might well be explained by stronger sociocultural norms regarding attractiveness toward girls than boys (Striegel-Moore et al., 2002), contributing to a stronger dissatisfaction with appearance in girls with a visible difference. However, it is important to remember that this gender effect was no longer significant when information about social experiences were included in the model. Another gender difference was that peer harassment mediated the association between cleft visibility and dissatisfaction with appearance only in girls. This finding indicates that a facial visibility may constitute a vulnerability factor for negative self-perceptions primarily in cases of harassment lending support to hypotheses of the impact of interpersonal experiences and sociocultural feedback on satisfaction with appearance. This is line with appearance evaluations equally mediating the association between social acceptance and lack of depressive symptoms in girls and boys.

Boys with a visible cleft did not report more positive ratings of appearance than the comparison group, in spite of all other variables indicating positive psychological functioning. One can speculate whether this indicates that satisfaction with appearance actually is being affected in this group, compared to perceptions of friendship, social acceptance, and emotional adjustment. This hypothesis would be consistent with more recent research, pointing to a growing recognition of the importance of body image and appearance also in boys and men (Tiggemann, Martins, & Churchett, 2008; Grammas & Schwartz, 2009; McCabe & Ricciardelli, 2004). The results thus would confirm the presence of risk related to appearance-concerns, and hence the interpretation of positive adjustment as an indication of emotional resilience.

6.7. Resilience or absence of risk?
Results from this study shed light on both risk and protective factors in adjusting to a facial difference, findings that should be discussed methodically to justify an interpretation of resilience. A methodological challenge is that risk factors tend to overlap. In the present area of research, dimensions of risk may include visibility/non-visibility, factors related to treatment and care, or be associated with other difficulties or syndromes. There is not enough knowledge about whether the effect of these dimensions is additive, or the manner in which
they might otherwise be combined to predict risk or psychological functioning (Lavigne et al., 1992)

Research in the area of resilience presents various methodological challenges. The preliminary criterion for resilience is its direct association to the presence of risk and adversity, which should be accurately specified. The presence of risk factors related to a cleft was presented in the first part of this study, and then further specified within each paper. In spite of obvious risk factors, the question remains whether the positive adjustments reported in this study are a result of active coping and resilience, or whether they indicate that a facial difference as a cleft actually does not constitute a risk factor.

One of the factors leading to resilience, the steeling effect of stress, actually show from appropriate exposure to risk, and the psychological and physiological coping mechanisms that are used in dealing with the adversity, resulting in hardiness (Karlsen, Dybdahl, & Vitterso, 2006; Rutter, 2009). Evidence suggests that a positive appraisal of the challenges associated with the cleft may moderate the deleterious effects of this condition (Edwards et al., 2005), pointing once again to the importance of self-perceptions and self-evaluations of adjustment. The results from this study may suggest that children and adolescents with a visible difference might have developed skills to cope with the consequences of the cleft. In addition, there are reasons to believe that efforts are made by the affected individuals and their families to strengthen positive aspects of friendships and social experiences, thus protecting the children and adolescents against the potential negative effects of a facial difference. Clinical experience also suggests that parents are heavily involved in their child’s treatment, and that they provide a supportive family atmosphere.

The discussion of whether it is appropriate to interpret positive outcomes as resilience in children with a cleft is an important question to address in further research. The question remains what differentiates children with visible differences who cope well from those with a visible difference who show a less positive adjustment. This study suggests that adolescents with a visible difference that feel socially confident can be expected to develop positive psychological well-being regardless of their actual popularity among peers, in line with previous findings (McElhaney et al., 2008). Results from this study suggest similar processes in appearance evaluations. Risk appears to be associated with subjective measures, while not being related to the actual objective presence of a facial difference. In other words, positive
self-evaluations of social acceptance, or self-assurance in one’s own appearance both act as a protective factor, whether appearance differs or not from the norms.

Resilience is not a static outcome, and will show ontogenetic instability. An optimal research design should be able to measure adjustment processes on repeated occasions, and hence measure the possible changes in how processes or variables operate as development proceeds. Future longitudinal research is necessary in order to study resilience in children and adolescents with a cleft lip and/or palate over time.

6.8. The importance of appearance evaluations

A longitudinal study specified that emotional distress resulting from media exposure to ideal body images was predicted by whether the girls showed preliminary vulnerability to negative body image (Stice, Spangler, & Agras, 2001). Hence, emotional distress in the presence of dissatisfaction with appearance when there is a facial difference may be mediated by the girls’ preliminary vulnerability to negative appearance evaluations.

The findings indicating that the objective measure of cleft visibility does not differentiate between children and adolescents that cope well compared to those who do not, lends support to empirical findings demonstrating the importance of the subjective ratings of appearance (Miyahara et al., 2006; Moss, 2005). This subjectively-perceived physical appearance is believed to reflect the individual’s perception of his or her visible difference (Kleve & Robinson, 1999), in line with the importance of cognitive appraisals in the determination of what constitutes stress (Lazarus & Folkman, 1984). It is the individual’s appraisal of whether the cleft constitutes a visible difference that has an impact on psychological adjustment, and not the objective existence of a visible difference. Accordingly, adolescents with a visible difference that feel socially confident and comfortable can be expected to develop positive psychological well-being regardless of their actual popularity among peers, as demonstrated by McElhaney et al. on adolescents without a facial difference (2008). The results of this study clearly confirm that research on visible differences must shift focus from the objective presence of a difference *per se*, to investigating the mechanisms behind subjective appearance satisfaction and factors which protect against or exacerbate appearance-related distress.

The concept of risk in the presence of a facial disfigurement is associated with the assumption that the individual’s physical characteristics may affect self-evaluations of appearance, in
addition to the challenges caused by a discrepancy between sociocultural norms of attractiveness, and potential negative interpersonal experiences. However, recent research indicates that appearance-concerns are increasing, reaching considerable proportions in the “normal population” (Lovegrove et al., 2005; Harris & Carr, 2001). The increasingly unrealistic standards of beauty may reinforce our appearance investment, one of the core attitudinal elements of body image schemas (Cash, 2002), or the importance an individual places on a specific domain of self-perception (Harter, 1999), leading to a more negative appearance evaluation. The importance individuals place on appearance in their life mediates the link between sociocultural factors and body dissatisfaction (Clark et al., 2007). In children and adolescents with a facial difference on the other hand, the more unattainable standard of “perfect appearance” may lead to a lower attitudinal investment in appearance, a re-definition of appearance as less important or central, hence protecting the individual against negative appearance evaluations. Children or adolescents with a facial difference may actively reduce the importance of appearance in their self-evaluations, in order to protect their overall self-perceptions. This has previously been illustrated in samples of girls, heavier adolescents appearing less concerned with conventional standards of attractiveness than girls with lower BMI (Dittmar et al., 2000). The present study was not aimed at testing Cash’s cognitive-behavioural model (Cash, 2002), or testing the validity of Harter’s model of development of self-perceptions (Harter, 1999). However, Cash’s model could guide the interpretations of the results. The fact that the physical characteristics of a facial difference does not predict self-evaluations of appearance directly is explained by the various developmental, cultural, and interpersonal experiences that also are part of the development of the self. Future research is needed in order to further confirm how these models may contribute in explaining adjustment to a visible difference.

6.9. Clinical implications: Treatment and prevention

Research which addresses the question “What makes a difference?” has the potential to expand our knowledge of how adversity processes function, contribute to more effective strategies to reduce risk and psychosocial maladjustment, providing intervention tools for the at-risk children. Further, such knowledge may facilitate preventive work. Consequently, parents of a newborn with a cleft could be given information about how they can develop the
coping repertoire of their child and turn the challenges of a cleft into a strengthening experience.

The present study was not aimed at developing treatment interventions. Hence, the results’ implications for prevention are only suggestive. Bearing this in mind, the results may suggest the necessity of preventive work directed toward children and adolescents at risk for appearance concerns, whether or not there is a visible difference. One of the factors shown as being related to psychosocial adjustment was the child’s emotional well-being, reminding us of the importance of a clinical screening concerning emotional health. Underlying emotional problems is probably an important factor with respect to the child’s adjustment to a visible condition. Further, results indicate a strong link between social experiences and subjective appearance evaluations, suggesting the possibility of strengthening social experiences in prevention and treatment of appearance-concerns. In view of the multiple components of adjustment that have been suggested in the present work, the importance of obtaining a more comprehensive picture of the child’s psychological functioning as an integral part of cleft care is reinforced.

Gender differences that were found in the adolescent sample confirm established risk factors for depressive symptoms, negative social experiences, and dissatisfaction with appearance in adolescent girls. These findings indicate that interventions should be specifically directed toward preadolescent girls, probably with a specific focus on girls with a visible cleft, in order to prevent the development of psychological distress.

Therapeutic interventions aimed at strengthening social competence have been demonstrated to be useful in the treatment of appearance concerns (Robinson et al., 1996), strengthening the assumption from the present study of the importance of social interaction in adjustment to a visible difference. Cognitive-behavioural interventions (Kleve, Rumsey, Wyn-Williams, & White, 2002) have also been demonstrated to improve social confidence and perceptions of social support. It is interesting that this intervention study also showed that participants perceived their disfigurement as less noticeable, results that support findings presented in this study, and interpretations related to self-appraisals and attitudinal dimensions of body image.

Research indicating a higher frequency of teasing when the cleft was visible suggests the importance of inquiring about experiences of harassment. Empirical findings indicating that the effect of early weight-teasing is mediated by early damage to the child’s well-being, in
addition to ongoing teasing (Eisenberg et al., 2006). Assessments of how the child copes emotionally with those negative experiences due to a visible difference are hence warranted. Equally important are interventions aimed at reducing teasing behaviours, and increasing protective factors such as friendships and social support.

7. Conclusion

The purpose of the present work was to explore the role of diverse social experiences, perceptions of appearance, and emotional well-being in adjusting to a visible difference as a cleft. Factors as gender, parental reports of teasing and emotional problems were also investigated, in addition to the presence of associated difficulties.

The results highlight that the objective measure of cleft visibility does not contribute to the explanation of the variation in psychosocial experiences or emotional well-being, in contrast to subjective measures. Primary findings show that a subjective appearance measure appears to act both as a potential risk and protective factor, depending on whether the social experiences are positive (friendships and social acceptance) or negative (harassment). In addition, appearance was shown to mediate the association between social acceptance and emotional adjustment in all groups of adolescents. Parental reports of teasing on the other hand, did not contribute to the explanation of psychosocial adjustment or subjective appearance satisfaction in the child, stressing the importance of assessing both the child’s and the parents’ perceptions of psychosocial experiences and emotional well-being. The results emphasize the importance of assessments of emotional distress in children, as depressive symptoms or anxiety. In addition, the present study suggests that a careful registration of all associated conditions is necessary in order to control for their potential effect on measures of psychological functioning.

The results also highlight possible gender differences in adjustment, indicating the presence of resistance factors in boys with a visible cleft, while possible vulnerability factors were found in adolescent girls with a visible cleft. However, in spite of possible vulnerability factors, the comparison of the clinical sample with a national reference group indicates that adolescents with a cleft cope well.

The importance of assessing the child’s and the adolescent’s subjective reports of satisfaction with appearance and psychosocial experience is in line with cognitive-behavioural models of
body image where objective measures of visibility are described as part of an individual’s physical characteristics, while the subjective appearance evaluation refers to the positive-to-negative evaluation of one’s appearance (Cash, 2002). This model contributes to explain the results from this study. Because physical characteristics are one among other developmental influences affecting appearance evaluations, the presence of a disfigurement can be expected to have less effect on self-evaluations than it would in isolation. Sociocultural factors and interpersonal experiences also influence appearance evaluations, reducing the impact of the physical characteristics that are associated with a disfigurement, reducing the importance of those characteristics, and strengthening self-representations. The results confirm that the processes involved in appearance-concerns are the same whether there is or not a facial difference. The consequences of a facial difference, hence, depend on the individual’s subjective response to that difference, and concurrent developmental and interpersonal experiences.
REFERENCES


