Risk and protective factors at age 16: Psychological adjustment in adolescents with a cleft lip and/or palate

Abstract

Objective: Explore psychological functioning in adolescents with a cleft at age 16 from a broad perspective, including cognitive, emotional, behavioural, appearance-related and psychosocial adjustment. High risk groups were identified within each area of adjustment, in order to investigate whether vulnerable adolescents were found across domains, or whether risk was limited to specific areas of adjustment.

Methods: Cross-sectional data based on psychological assessments at age 16 (*n* =857) for adolescents with CLP were obtained and compared to data from a large reference group and/or norms. The effect of gender, cleft visibility and the presence of an additional condition were investigated on all outcome variables.

Measures: Hopkins Symptom Checklist, Harter Self-Perception Profile for Adolescents, Child Experience Questionnaire, Strengths and Difficulties Questionnaire, Satisfaction with Appearance scale.

Results: The main factor influencing psychological adjustment across domains was gender, with girls in general reporting more psychological problems, as is seen in reference groups. The presence of an additional condition also affected some of the measures negatively. No support was found for cleft visibility as a risk factor except for dissatisfaction with appearance. Correlation analyses of risk groups seem to point to an association between social and emotional risk, and between social risk and dissatisfaction with appearance, while associations between other domains were found to be weak.

Conclusions: The results point to areas of both risks and strengths in adolescents born with a cleft lip and/or palate. Future research should investigate how protective factors could counteract potential risk in adolescents with a cleft.

Key Words: Visible difference; cleft lip and palate; psychosocial adjustment; cognitive function; emotional adjustment; satisfaction with appearance; behaviour.

Introduction

Research exploring the psychological impact of cleft lip and/or palate (CL/P) currently provides an inconsistent picture of how individuals adjust to this condition. Adolescence is understood to be a difficult period for all young people, since the desire to qualify as 'normal' forms a central part of wellbeing at this time (Nurmi, 2004). Associations between social and emotional risk and dissatisfaction with appearance during adolescence have been well documented within the general population (Paxton et al., 2006; Dekker et al., 2007). Given that physical appearance is often the prime focus for evaluating whether an individual 'blends in' with a peer group, and that more emphasis is placed on appearance as the gateway to forming friendships and romantic relationships during adolescence (Rumsey and Harcourt, 2005), having CL/P may become particularly problematic for young people at this developmental stage.

Adolescents with CL/P may be at risk in a number of adjustment domains, including social experiences, dissatisfaction with appearance, behavioural difficulties, cognitive function, and emotional distress (Hunt et al,. 2005; Collett and Speltz, 2007). However, some studies have found that young people with CL/P do not experience psychosocial difficulties above that experienced by the general population (Locker et al., 2005; Pope and Snyder, 2005; Berger and Dalton, 2009). Recent research has also reported a number of positive findings among young people with a cleft, such as positive coping strategies, stress-related growth, and more positive perceptions of social experiences, which are indicative of protective factors and processes of resilience (Baker et al., 2009; Berger and Dalton, 2009; Feragen et al., 2010).

Such findings may be suggestive of different domains of risk and resilience working within the same individual, with adolescents with CL/P demonstrating good adjustment in some areas while being at risk in others, as has been shown within the general resilience literature (Kumpfer, 1999; Masten, 2001; Luthar, 2006). Although some studies in the field of CL/P have investigated potential associations between different areas of adjustment (Berger and Dalton, 2011), few studies have looked at adjustment across more than one domain. This also makes it difficult to compare risk groups across measures. Information regarding specific or potential risk and protective factors could help primary care providers and cleft teams to more accurately target those who may need more intensive care, while also capitalising on existing strengths and fostering resilience. To our knowledge, no studies to date have specifically aimed to explore both risk and protective factors within the same study.

Inconsistent findings are almost certainly indicative of the multifaceted and complex processes involved in psychological adjustment, but may also be partially explained by methodological issues, such as a wide range of study designs, difficulties accessing large and representative samples, and considerable variation in the choice of psychological instruments used (Rumsey and Stock, 2013). Such methodological limitations make it difficult to compare findings across studies and to discriminate between those adolescents who cope well and those who may be at risk of psychological difficulties. To capture profiles of adjustment in adolescents with CL/P effectively, there is a need to agree upon measures which are both clinically useful and psychometrically robust. Unfortunately, the process of choosing such measures is challenging, and dialogue among researchers and clinicians is ongoing. A question has also been raised as to whether generic measures of psychological wellbeing, condition-specific measures, or a combination of the two are most helpful (Rumsey and Stock, 2013).

A second methodological concern is the presence of several additional conditions known to impact on psychological functioning within CL/P samples. Although in the past researchers have often excluded those with a diagnosed syndrome from their analyses, other milder conditions such as attention deficit and/or hyperactivity disorder and developmental delay, may still be present. As well as potentially affecting a study's conclusions, little is learned about those individuals who have been excluded. Given the high prevalence of additional conditions in children born with a cleft (Milerad et al., 1997; Swanenburg et al., 2003; Feragen et al., in press), and the recent finding that those with an additional condition may be at higher psychological risk (Feragen and Stock, 2014), any additional conditions within a sample need to be included, identified and categorised accordingly.

Other additional factors warranting further investigation include gender and cleft visibility. Within the general population, females are thought to be much more likely to experience dissatisfaction with appearance (Striegel-Moore and Franko, 2002). Additionally, appearance dissatisfaction appears to be more closely linked to self-esteem in females, contributing considerably to emotional distress (Nolen-Hoeksema and Girgus, 1994). However, females in the general population often report having more numerous and more intimate friendships than their male peers (Galambos, 2004). For boys, elevations in behavioural difficulties and peer problems have been observed (Goodman, 1997; Van Roy et al., 2006). Similar gender patterns have been implicated to some degree in adolescents with CL/P (Berger and Dalton, 2009; Feragen et al., 2010). However, findings within the CL/P population as a whole are conflicting, and it is unclear whether the observed gender differences are a reflection of normal differences within the general population (Berger and Dalton, 2011), or whether gender could be a moderating factor for risk and protection in adolescents with a cleft. A second debate within the cleft literature is the degree to which the objective visibility of a cleft impacts on adjustment. While some studies have suggested that an individual's

subjective perceptions of their appearance, or self-perceptions, outweigh the severity of a visible difference (Moss, 2005; Appearance Research Collaboration, 2009; Feragen et al., 2010), many studies continue to investigate cleft visibility as a key variable, with some significant findings (Berger and Dalton, 2009; Mani et al., 2013; Millar et al, 2013). More generally, some differences in adjustment have been identified in relation to cleft type. For example, those with palatal involvement have been shown to have greater or differing cognitive problems than those with other cleft diagnoses, and when compared to their peers from matched comparison groups (Speltz et al., 2000; Millard and Richman, 2001; Roberts et al., 2012).

To investigate many of the above issues, large and representative samples are needed. In addition, if control groups or reference groups are not included in studies, then the conclusions drawn about gender and other variables are less meaningful. Since more boys than girls have a visible cleft, reported differences between cleft types could be associated with gender differences. Some differences could also be due to age, since studies often include children who are at differing developmental stages in their samples. Large samples, discussed in the context of comparable reference groups, would make it possible to investigate possible interactions between potentially interesting variables.

The first aim of the present study was to explore adjustment across a range of domains that are considered central during childhood and adolescence, and that have been shown to be important in cleft research. These five domains were: cognitive, behavioural, emotional and social functioning, and satisfaction with appearance. The effects of gender, cleft visibility and the presence of an additional condition on the outcome variables were evaluated individually, in addition to possible interactions between them. A secondary aim was to identify a high risk group within each domain, in order to investigate whether risk factors co-varied across groups, or whether risk was restricted to specific domains of adjustment. This approach made

it possible to investigate whether risk could be general or domain-specific, hence indicating positive adjustment in the remaining adjustment domains within the same individual. To the authors' knowledge, this is one of the first papers to include such a wide range of domains across a large sample, and to explore both risk and protective factors within a single study. A final aim was to present and discuss psychometric properties in relation to each outcome variable.

Method

Setting

The Oslo-team is responsible for two thirds of the entire Norwegian cleft population, treating approximately 70-80 new babies born with a cleft per year. The present study was based on data from case records of 16-year-old adolescents with cleft lip and/or palate. The team's clinical psychologist briefly met with each participant before he or she completed a questionnaire that included the measures used in the present study. One of the measures was also provided by each adolescent's parent(s).

Participants

Adolescents

The aim of the present study was to ascertain the methodological strengths and weaknesses of the measures used in routine clinical assessment, and their ability to identify risk groups. Hence, data were based on clinical audit. Patient confidentiality was preserved, and the Regional Committee for Medical Research Ethics granted ethical approval for the study. All adolescents ($n = 857$) who attended the routine 16-year-old follow-up from January 2004 to December 2013 were eligible for inclusion in the study, hence 10 consecutive birth cohorts. No participants were excluded from the study. However, due to severe developmental

problems, some adolescents (n= 70) were not able to attend the routine assessments, and outcome measures are missing for this group.

In the cleft sample, 329 adolescents were female and 518 adolescents were male. Adolescents' cleft type included cleft lip and palate, CLP ($n = 310$), cleft lip or cleft lip alveolus, CLA ($n = 253$), cleft palate, CP or submucous cleft palate, SMCP ($n = 284$). Information about gender and cleft type was missing for ten adolescents. For the purpose of the statistical analyses, the adolescents were categorized into two groups: adolescents with visible clefts (CLP and CLA, $n = 563$) and adolescents with non-visible clefts (CP/SMCP, $n = 284$). Among the girls, 47.7% had a non-visible cleft and 52.3% had a visible cleft. Among the boys, 24.5% had a non-visible cleft and 75.5% had a visible cleft. Some of the children and adolescents within the samples were of non-Caucasian origin ($n = 83/769$; 9.7%), and 52/809 were adopted (6.4% of the total sample).

When available, scores from the adolescent cleft sample were compared to a large reference group consisting of adolescents 16 years–olds ($n = 1832$; 888 girls, 944 boys) who completed a national representative survey ''Young in Norway 2002'' with a participation rate of 92.3%. The reference group completed the Self-Perception Profile for Adolescents (except the Romantic Appeal subscale) and the Hopkins Symptoms Checklist. Perceptions of Romantic Appeal were compared to Norwegian norms (Wichstrøm, 1998). Results from the Strengths and Difficulties Questionnaire were compared to large samples of same-aged Norwegian adolescents (Rønning et al., 2004; Van Roy et al., 2006). Parent data were not available for the reference groups.

Parents

The Strengths and Difficulties Questionnaire (SDQ) was included in routine assessments from 2010 onwards. Hence, parent reports were only available for the last four birth cohorts ($n =$

304). Among the parents, 51.3% were mothers ($n = 156$), 22.4% fathers ($n = 68$), and 24.3% were completed by both parents ($n = 74$). Two percent were not the child's parents ($n = 6$), and included siblings, grandparents and foster parents.

Additional conditions

Information was gathered about additional diagnoses or conditions which have been associated with cognitive and/or psychological functioning, such as developmental delay, learning difficulties, dyslexia, autism spectrum disorders and attention deficit and/or hyperactivity disorders (AD/HD). A total of 26.3% of the adolescents ($n = 225$) had one or several additional conditions: developmental delay/difficulties (7.4%), dyslexia or specific language difficulties (7.7%), autism spectrum disorder (2.1%), or AD/HD (7.5%). Furthermore, some adolescents had a diagnosed syndrome (7.4%), such as 22Q11.2, Treacher Collins, or Goldenhar, with or without associated psychological and/or cognitive difficulties. While 52.9% of the 225 adolescents with an additional condition had one diagnosed condition in addition to the cleft, 24.9% had two additional diagnoses, while the remaining 22.2% had three or more conditions in addition to the cleft. Information about additional conditions was found in the child's case records, and/or was given by the adolescent's parents.

Measures and Instruments

Childhood Experience Questionnaire (CEQ)

The CEQ (Pertschuk & Whitaker, 1982) reflects the child's self-reporting of social experiences on a 5-point Likert scale. The measure includes 20 items which relate to topics such as relationships with friends ("I spend time with friends at school"), social isolation ("I try to hide from people", "I keep to myself"), and involvement in new experiences ("I go to new places"). 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 possess satisfactory internal consistency and a coherent factor structure (Emerson, Spencer-Bowdage, & Bates, 2004).

Strengths and Difficulties Questionnaire (SDQ)

The SDQ (Goodman, 1997) is a screening tool for behavioural difficulties and strengths in children and adolescents, and was completed by both the parent(s) and the adolescents in the current study. The SDQ has been used systematically at psychological routine assessment at age 16 in the Oslo-team since the end of 2009. Data therefore exists for more than three consecutive birth cohorts, providing both self-reported and parent-reported data. The SDQ includes five subscales measuring emotional distress, conduct problems, hyperactivity and/or attention difficulties, peer relationship problems and pro-social behaviour. Each subscale includes five items that are positively or negatively worded. Each of the 25 items is scored "not true", "somewhat true" or "certainly true" (0-2). The first four subscales are summarized into the total difficulties score (20 items in total, with a total score ranging from 0-40). Cut-off points for identifying adolescents at risk are recommended to be at the 90th percentile. The cut-off scores presented by Goodman (www.sdq.info) have been slightly adjusted to a Norwegian population and are the ones used as a reference in the present study (Van Roy et al., 2006).

Satisfaction With Appearance Scale (SWA)

The SWA (developed by the Psychology Special Interest Group of the Craniofacial Society of Great Britain and Ireland) reflects satisfaction with cleft-related and non-cleft-related parts of the face, speech, overall appearance and the perceived visibility of the 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 4 item version of the scale was used in the present study, measuring satisfaction with overall appearance and with the face, in addition to the adolescent's perception of cleft visibility and effect on social relationships. The SWA has been reported to possess satisfactory internal consistency and a coherent factor structure (Emerson et al., 2004). The mean total score of the 4 items were used (Range 0-10).

Self-Perception Profile for Adolescents (SPPA)

The SPPA (Harter, 1988) is a 35 item scale designed to assess adolescents' self-perceptions in a number of psychological domains. The Norwegian version was used, which has been shown to achieve better reliability, better convergent validity, and a replication of the same factorial pattern than the original version (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. The following subscales were administered: Scholastic Competence, Social Acceptance, Physical Appearance, Romantic Appeal, Close Friendships and Global Self-Worth.

Hopkins Symptom Checklist (HSCL-25)

The shortened version of the HSCL-25 (Kandell & Davies, 1982) measures depressive symptoms through 7 items. Tambs and Moum (1993) have demonstrated that this strongly abbreviated version of the instrument correlates well (r = .92) with the original HSCL-25. Each item, such as "Feeling unhappy, sad, or depressed" and "Feeling hopeless about the future", was rated on a frequency of occurrence over the preceding 14 days, ranging from "Never" (1) to "Very much" (4). Mean scores were calculated. Severity of symptoms were categorized using cut-off scores where mean scores below 1.75 indicate no or mild frequency

of symptoms, values between 1.75 and 2.00 indicate moderate problems and mean scores above 2.00 indicate severe symptoms (Rognerud et al., 2002).

Statistical Method

SPSS 21 was employed for the statistical analyses. Calculations of internal reliability for all subscales are provided in the first section of the results. Concurrent validity was explored by calculating Pearson's correlation values between subscales that measure similar dimensions, across measures and across informants (adolescents and parents). The second part of the results investigated the outcome variables according to the study's aims, and the identification of high risk groups. In order to enhance readability, the second part of the results is presented in the following order for each outcome variable:

i.   A $2 \times 2 \times 2$ ANOVA exploring the main effects and potential interactions of gender, cleft visibility and the presence of an additional condition on the outcome variable. The ANOVA test provides adjusted effects of means and avoids an accumulation of Type I errors as would be the case with successive t-tests. Eta square (η^2) effect sizes were calculated. Cohen's guidelines (1988) were used to interpret η^2: small effect: 0.01; medium effect: 0.059; large effect: 0.138. Effect sizes were not calculated in cases of statistically non-significant results.

ii.   Comparisons between the cleft sample and reference groups/norms and/or clinical cut-off scores. Reference groups were compared to adolescents with a cleft and no additional condition. Mean scores (M) and Standard deviations (SD) were compared according to gender between the cleft sample and the reference group. Mean scores for cleft types are only presented separately in cases of significant main effects or interactions involving cleft visibility.

When comparing several subgroups, Tukey post hoc multiple-comparisons tests were used, with corresponding mean difference (MD) and standard error (SE). Calculations of effect size were performed using Cohen's *d.*

iii.   Identification of a high risk group according to norms (for the SDQ and SCL-7) or according to scores below the 10th percentile (CEQ, SWA and SPPA). A dichotomous variable was created in order to explore the characteristics of the high risk groups with respect to gender, cleft visibility, and the presence or absence of an additional condition. Chi-square analyses were used when investigating differences between the categorical variables.

Last, five new variables were created, classifying risk according to five different domains of adjustment (cognitive, behavioural, social, emotional, and appearance-related), irrespective of which outcome variables that had been used in the first instance. In addition to the identified high risk group, borderline cases were also identified by using provided cut-off scores (SDQ) or by calculating cut-off scores below the 25th percentile (SPPA, CEQ and SWA). Cut-off scores were calculated based on the reference group for the SPPA. In order to investigate a potential co-variation between the risk groups, Pearson's correlation coefficient was used.

Results

Psychometric properties of the outcome measures

The SPPA, HSCL-7*,* and SDQ are all validated measures, while the CEQ and the SWA are not. Internal reliability was calculated for all measures and is reported in Table 1. As can be seen from the table, the psychometric properties varied significantly across and within measures, and irrespective of whether they have been validated in the past or not. While all subscales from the SPPA, the HSCL-7*,* the SWA, and the CEQ had acceptable to excellent internal reliability, reliability was more questionable for self-reports of problems with

attention and/or hyperactivity (SDQ), and self- and parent reports of peer problems (SDQ). The subscale measuring conduct problems had unacceptable reliability.

General adjustment and self-perceptions

General adjustment was measured using the Global Self-Worth scale of the SPPA and the Total Score of the SDQ (self- and parent reports). Correlations between the measures used are provided in Table 2.

Global Self-Worth (SPPA)

2×2×2 ANOVA: One three-way interaction and one main effect were found on the Global Self-Worth scale (Table 3). Girls with a non-visible cleft and no additional condition reported more positive perceptions (EMM = 3.0, SE = .06) than in cases of a visible cleft (EMM = 2.8, SE = .09). In boys, the opposite was the case (CP: EMM = 3.3, SE = .07; CLP: EMM = 3.4, SE = .04). In adolescents with an additional condition, there were no differences in means between the two groups of girls (CP: EMM = 2.9, SE = .05; CLP: EMM = 2.9, SE = .12), while boys with CLP reported somewhat more negative self-perceptions (EMM = 3.3, SE = .08) than boys with CP (EMM = 3.4, SE = .11). The effect size for this three-way interaction was however small (η^2 = 0.0045). The main effect was related to gender, with girls reporting lower global self-perceptions (EMM = 2.9, SE = .04) than boys (EMM = 3.4, SE = .04; F (1,702) = 71.51, *p* < .001). Effect size was medium to large (η^2 = 0.083). There were no differences relating to cleft visibility (F (1,702) = 0.69 *p* > .05) or the presence of an additional condition (F (1,702) = 0.00, *p* > .05).

Reference group comparisons: Girls with a visible cleft reported similar self-perceptions (M = 2.8, SD = .71) as girls from the reference group (M = 2.8, SD = .68; MD = .02, SE = .06, *p* > .05), while girls with a non-visible cleft reported slightly more positive self-perceptions (M =

2.9, SD = .70). However, calculations of Cohen's *d* revealed that effect sizes were small ($<$ 0.2). Boys with a visible cleft, reported significantly higher global self-perceptions (M = 3.4, SD = .50) than boys from the reference group (M = 3.1, SD = .59; MD = .26, SE = .04, *p* $<$ .001, *d* = 0.48). This was also the case for the boys with a non-visible cleft (M = 3.3, SD = .53, MD = .18, SE = .06, *p* $<$ .01; *d* = 0.32). The difference between boys with a non-visible and a visible cleft was not statistically significant (MD = -.08, SE = .07, *p* $>$ .05).

High risk group analysis: In order to explore high risk groups, a percentile analysis was performed on the reference group, revealing that a mean of 2.2 or lower was indicative of very low perceptions of global self-worth ($< 10^{th}$ percentile). A dichotomous variable was created, identifying the adolescents at high risk and exploring how gender, cleft visibility and the presence of an additional condition varied in the high risk group. There were significantly more girls in the high risk group, 22.1% (*n* = 61) compared to 2.8% of boys (*n* = 12, χ^2 = 67.81, *p* $<$ .001). The reference group had the same frequency of girls in the high risk group (24%), while more boys were at high risk (9%) in the reference group compared to the cleft sample. There were no differences related to cleft visibility (χ^2 = 1.67, *p* $>$ .05) or the presence of an additional condition (χ^2 = 0.28, *p* $>$ .05).

Total Score (SDQ)

2×2×2 ANOVA: No interactions were found in self-reports or in parent reports (Table 3). Two main effects were found in self-reports, with girls reporting more general problems (EMM = 11.6, SE = .50) than boys (EMM = 9.1, SE = .46; F (1,327) = 13.70, *p* $<$ .001; η^2 = 0.038), and adolescents with a cleft only reporting less problems (EMM = 9.6, SE = .38) than adolescents with an additional condition (EMM = 11.1, SE = .55; F (1,327) = 4.64, *p* $<$ .05; η^2 = 0.013). Two main effects were found on parent reports, the most important of which related to the presence of an additional condition (EMM = 5.7, SE = .45, EMM = 10.1, SE = .62; F (1,290)

= 32.77, $p < .001$; $\eta^2 = 0.090$). According to parents, girls had more general problems (EMM = 8.7, SE = .57) than boys (EMM = 7.1, SE = .51; $F(1,290) = 4.40$, $p < .05$; $\eta^2 = 0.011$).

Reference group comparisons: Compared to the reference group, girls with a cleft and no additional condition reported the same level of problems (M = 10.7, SD = 5.72) as girls from the reference group (M = 10.6, SD = 5.51), while boys with a cleft (M = 8.2, SD = 4.25) reported less problems than boys from the reference group (M = 10.2, SD = 5.35; $d = 0.41$).

High risk group analysis: According to self-reports, 22 adolescents (6.7%) were identified as being at high risk of psychological difficulties, and 24 (8.2%) according to the parent reports. Self-reports identified more girls (12.1%, $n = 15$) than boys (2.4%, $n = 7$; $\chi^2 = 9.33$, $p < .01$), while there were no differences related to cleft visibility ($\chi^2 = 2.61$, $p > .05$). According to parent reports, there were more adolescents with CP at risk (16.1%) than in cases of a visible cleft (4.9%; $\chi^2 = 10.09$, $p < .01$). The presence of an additional condition also represented a risk factor according to self-reports (11.3% vs. 4.7%; $\chi^2 = 4778$, $p < .05$) and parent reports (17.4% vs. 4%; $\chi^2 = 14.98$, $p < .001$).

Cognitive Function

Cognitive function was measured by the Scholastic Competence scale of the SPPA and the Attention/Hyperactivity scale of the SDQ (self- and parent reports). Correlations between the measures used are provided in Table 2.

Scholastic Competence (SPPA)

2×2×2 ANOVA: No interactions and two main effects were found. These are reported in Table 3. Girls reported lower scholastic competence (EMM = 2.8, SE = .04) than boys (EMM = 2.9, SE = .04; $F(1,702) = 7.12$, $p < .05$). Effect size was small ($\eta^2 = 0.008$). The other main effect was that adolescents with an additional condition reported lower scholastic competence

(EMM = 2.7, SE = .05) than adolescents without an additional condition (EMM = 3.0, SE = .03; F (1,702) = 16.53, $p < .001$; $\eta^2 = 0.021$).

Reference group comparisons: Compared to the reference group, girls with a cleft (M = 2.9, SD = .66) reported similar perceptions of Scholastic Competence (M = 2.9, SD = .56; F (1,1163) = .05, $p > .05$). The difference in means between the two groups of boys were statistically significant (cleft: M = 3.0, SD = .56 vs. reference group: M = 2.9, SD = .55, F (1,1375) = 3.89, $p < .05$), although effect size was small ($d = 0.18$).

High risk group analysis: A mean of 2.20 or lower ($< 10^{th}$ percentile) was indicative of very low perceptions of Scholastic Competence in the reference group. A total of 98 adolescents with a cleft (14.1%) were found to be at high risk. There were 18.6% girls ($n = 51$) compared to 11.2% boys ($n = 47$; $\chi^2 = 7.60$, $p < .01$). There were no differences related to cleft type ($\chi^2 = .16$, $p > .05$). In addition, a higher proportion of adolescents with a cleft and an additional condition were found in the high risk group (21.4%, $n = 33$) than a cleft only (12.0%, $n = 65$; $\chi^2 = 8.77$, $p < .01$). The same percentage of adolescents was at high risk for low perceptions of Scholastic Competence in the cleft sample (14.1%) as in the reference group (14.1%).

Attention and Hyperactivity (SDQ)

2×2×2 ANOVA: No interactions were found in either self-reports or parent reports (Table 3). There was one main effect, with more problems with attention and hyperactivity in adolescents with an additional condition than in the young people with a cleft only in self-reports (EMM = 3.8, SE = .17 vs. EMM = 4.5, SE = .25; F (1, 327) = 6.47, $p < .05$; $\eta^2 = 0.020$) and in parent reports (EMM = 2.0, SE = .19 vs. EMM = 4.0, SE = .26, F (1, 290) = 37.41, $p < .001$; $\eta^2 = 0.102$). As can be seen in Table 3, no main effects were found relating to gender or cleft visibility.

Reference group comparisons: Compared to the reference group, girls with a cleft and no additional condition reported less problems with attention and hyperactivity (M = 3.8, SD = 2.36) than girls from the reference group (M = 4.1, SD = 3.43), as was the case for the boys with a cleft (M = 3.8, SD = 2.11) when compared to the boys from the reference group (M = 4.0, SD = 2.30). However, effect sizes were small ($d < 0.2$).

High risk group analysis: Cut-off points revealed 21 adolescents (6.4%) within the high risk group according to self-reports, while parents identified 30 adolescents (10.3%). Neither gender, cleft visibility nor the presence of an additional condition varied significantly in the high risk group on self-reports (χ^2 = .94, .03, and 3.55, all p > .05). In parent reports, there were no gender differences (χ^2 = .10, p > .05). However, there were more adolescents with a non-visible cleft (18.4%, n = 16) than with a visible cleft (6.9%, n = 14; χ^2 = 8.77, p < .01) in the high risk group, and more adolescents with a cleft and an additional condition (23.9%, n = 22) than a cleft only (4.0%, n = 8; χ^2 = 27.11, p < .001).

<div align="center">

Behavioural difficulties

</div>

Behavioural difficulties were measured by the Conduct Problems subscale on the SDQ (self- and parent reports). Correlations between the measures used are provided in Table 2.

Conduct problems (SDQ)

No interactions and no main effects were found in self-reports (Table 3). In parent reports, there were two 2-way interactions and one main effect. The interactions pointed to more conduct problems in girls with an additional condition (EMM = 1.8, SE = .20) when compared to girls with a cleft only (EMM = 1.1, SE = .15), while there were no differences between the two groups of boys (EMM = .9, SE = .18 and EMM = 1.0, SE = .13; F (1,290) = 6.87, p < .01). The second interaction was between cleft visibility and the presence of an additional condition. Adolescents with a CLP and an additional condition had more conduct

problems (EMM = 1.5, SE = .19) than in cases of a CLP only (EMM = .8, SE = .10), while

the impact of an additional condition was not significant in cases of a CP (EMM = 1.3, SE =

.17 and EMM = 1.2, SE = .20; F(1, 290) = 4.79, $p < .05$). Both interactions had small effect

sizes ($\eta^2 < 0.02$). The main effect was related to gender and suggested that girls had more

conduct problems (EMM = 1.5, SE = .13) than boys (EMM = .9, SE = .11; F(1,290) = 10.58,

$p < .01$; $\eta^2 = 0.030$). This could be due to the impact of an additional condition in the group of

girls as reported in the two-way interaction.

Reference group comparisons: Girls with a cleft reported less conduct problems (M = 1.4, SD

= 1.37) than girls from the reference group (M = 1.8, SD = 1.65; $d = 0.26$). The difference

was even greater in the group of boys (M = 1.4, SD = 1.26 and M = 2.3, SD = 1.88; $d = 0.56$).

High risk group analysis: Self-reports revealed 10 adolescents (3.0%) within the high risk

group, and 12 according to parent reports (4.1%). Neither gender, cleft visibility nor the

presence of an additional condition varied significantly in the high risk group on self-reports

($\chi^2 = .67$, .47, and .55, all $p > .05$). Parents reported more conduct problems in girls (8.3%, $n =$

9) than in boys (1.6%, $n = 3$; $\chi^2 = 7.59$, $p < .05$), and no differences related to cleft visibility

($\chi^2 = .83$, $p > .05$), nor to the presence of an additional condition ($\chi^2 = .02$, $p > .05$).

Social Experiences, Social Acceptance and Friendships

Social experiences were measured by the Child Experience Questionnaire of the CEQ, the

Peer Problems subscale of the SDQ, the Social Acceptance, Close Friendships and the

Romantic Appeal subscales of the SPPA. Correlations between the measures used are

provided in Table 2.

Social experiences (CEQ)

2×2×2 ANOVA: There were no interactions and one main effect (Table 3). The only main effect was gender, with girls reporting less positive social experiences (EMM = 2.6, SE = .04) than boys (EMM = 2.8, SE = .04; F (1,488) = 12.51, $p < .001$; $\eta^2 = 0.023$). As can be seen in Table 3, there were no main effects relating to cleft visibility (F (1,488) = 1.00, $p > .05$) or to the presence of an additional condition (F (1,488) = 2.56, $p > .05$).

Lack of reference group: As far as we know, no norms exist for the CEQ, and no studies have provided a reference group that would make comparisons with the current sample possible.

High risk group analysis: Percentile analyses revealed that a mean of 2.10 or lower was indicative of high psychosocial risk. A total of 57 adolescents (11.6%) were identified at high risk of negative social experiences. There were more girls (16.5%, $n = 32$) than boys at high risk (8.4%, $n = 25$; $\chi^2 = 7.39$, $p < .01$), and the presence of an additional condition was also a risk factor (18.0%, $n = 24$ vs. 9.2%, $n = 33$; $\chi^2 = 7.30$, $p < .05$). There were no differences relating to cleft visibility ($\chi^2 = 1.81$, $p > .05$).

Peer problems (SDQ)

2×2×2 ANOVA: One three way interaction was found, revealing that, in cases of a cleft only, girls with CLP reported more peer problems on self-reports (EMM = 1.9, SE = .21) than girls with CP (EMM = 1.6, SE =.28), while the opposite was the case for the boys (EMM = 1.2, SE = .14 and EMM = 2.1, SE = .29). When there was an additional condition, girls with CP reported more peer problems (EMM = 2.1, SE =.31) than girls with CLP (EMM = 1.5, SE =.40), while the difference in the boys seemed less important (EMM = 1.4, SE =.40 and EMM = 1.6, SE =.24; F(327) = 5.68, $p < .05$). Effect size for the interaction was small ($\eta^2 = 0.015$). There were no main effects on self-reports (Table 3). In parent reports, there were no interactions and one main effect, with parents reporting more peer problems in adolescents

with an additional condition (EMM = 2.3, SE = .21) than in cases of a cleft only (EMM = 1.3, SE =.15; F (290) = 13.37, $p < .001$; $\eta^2 = 0.040$).

Reference group comparisons: Girls with a visible cleft and no additional condition reported somewhat more peer problems (M = 1.9, SD = 1.62) than girls from the reference group (M = 1.6, SD = 1.56) and girls with a non-visible cleft (M = 1.6, SD = 1.85). However, effect sizes were small ($d < .02$). Boys with a non-visible cleft and no additional condition reported similar levels of peer problems (M = 2.1, SD = 1.90) as boys from the reference group (M = 2.0, SD = 1.84), while boys with a visible cleft reported significantly less peer problems (M = 1.2, SD = 1.25). Effect size was moderate ($d = 0.51$).

High risk group analysis: Cut-off points revealed 19 adolescents (5.8%) within the high risk group according to self-reports, and 41 (14.0%) according to parent reports. Self-reports identified more girls (10.5%, $n = 13$) than boys (2.9%, $n = 6$; $\chi^2 = 8.11$, $p < .01$) in the high risk group, while parent reports did not point to gender differences ($\chi^2 = 2.67$, $p > .05$). There were no differences relating to cleft type in self-reports ($\chi^2 = 2.83$, $p < .01$), while parent reports identified more cases of CP (25.3%, $n = 22$) than CLP (9.3%, $n = 19$; $\chi^2 = 12.86$, $p < .01$). The presence of an additional condition was not associated with social risk according to self-reports ($\chi^2 = 1.55$, $p > .05$), while parents reported a higher frequency of adolescents with social problems when the cleft was associated with an additional condition (9.5%, $n = 19$ vs. 23.9%, $n = 22$; $\chi^2 = 10.85$, $p < .01$).

Social acceptance (SPPA)

2×2×2 ANOVA: There were no interactions and one main effect, related to gender (Table 3). Girls with a cleft reported lower social acceptance (EMM = 3.1, SE = .04) than boys (EMM = 3.3, SE = .04; F (1,702) = 14.31, $p < .001$; $\eta^2 = 0.019$). Cleft visibility and the presence of an additional condition were not significant (F (1,702) = 0.30 and 2.18, $p > .05$).

Reference group comparisons: Compared to the reference group, girls reported similar self-perceptions of Social Acceptance whether they had a cleft (M = 3.1, SD = .64) or were from the reference group (M = 3.1, SD = .54; F (1, 1162) = 1.56, p > .05). The difference between the two groups of boys was statistically significant (Cleft group: M = 3.3, SD = .56; Reference group: M = 3.2, SD = .54; F (1, 1374) = 12.62, p < .001; d = 0.18).

High risk group analysis: Percentile analyses revealed that a mean of 2.40 or lower was indicative of high risk in the reference group (n = 190, 10.4%). A dichotomous variable was created, identifying 69 adolescents with a cleft at high risk (9.8%). There were more girls at high risk (15.6%, n = 49) than boys (6.0%, n = 26; χ^2 = 17.41, p < .001) in the cleft sample, while there was no gender difference in the reference group (χ^2 = .58, p > .05). There were no differences relating to cleft visibility (χ^2 = .74, p > .05), and no differences relating to an additional condition (χ^2 = 2.03, p > .05).

Close Friendships (SPPA)

2×2×2 ANOVA: A three way interaction was found, in addition to one main effect. In cases of a cleft only, girls with a non-visible cleft reported somewhat more positive perceptions of close friendships (EMM = 3.5, SE = .05) than girls with a visible cleft (EMM = 3.4, SE = .05), while the opposite was the case in the group of boys (CP: EMM = 3.2, SE = .07; CLP: EMM = 3.5, SE = .03). When there was an additional condition, boys and girls with a visible cleft reported somewhat more positive perceptions of close friendships (EMM = 3.4, SE = .07 and EMM = 3.5, SE = .11) than boys and girls with a non-visible cleft (EMM = 3.3, SE = .10 and EMM = 3.3, SE = .09; F(1, 702) = 4.45, p < .05). However, effect size for the interaction was small (η^2 = 0.006). The only significant main effect was related to cleft visibility, with adolescents with a visible cleft reporting more positive perceptions of close friendships (EMM = 3.4, SE = .04) than adolescents with a non-visible cleft (EMM = 3.3, SE = .04; F

$(1,702) = 4.16$, $p < .05$; $\eta^2 = 0.005$). There were no differences relating to gender (F $(1,702) =$ 1.80, $p > .05$) or an additional condition (F $(1,702) = .36$, $p > .05$).

Reference group comparisons: Since the 2×2×2 ANOVA revealed a main effect of gender and cleft visibility, means were compared across all four groups. There were no differences in means between the three groups of girls (CP: M = 3.4, SD = .55; CLP: M = 3.4, SD = .57; Ref.gr.: M = 3.4, SD = .56). However, differences did appear between the groups of boys, where boys with a visible cleft reported more positive perceptions of close friendships (M = 3.4, SD = .53) than boys with a non-visible cleft (M = 3.3, SD = .62; MD = .11, SE = .06, $p >$ .05), and more positive perceptions than the boys from the reference group (M = 3.1, SD = .59; MD = .30, SE = .04, $p < .001$; $d = 0.54$). Boys with a non-visible cleft also reported more positive self-perceptions of close friendships than the reference group (MD = -.18, SE = .07, $p < .05$; $d = 0.33$).

High risk group analysis: A high risk group of 51 adolescents (7.4%) was identified as having mean scores below 2.40, as compared to 187 adolescents (10.2%) in the reference group. There were no variations due to gender ($\chi^2 = .85$, $p > .05$), no effects of cleft visibility ($\chi^2 = .85$, $p > .05$) and no differences relating to the presence of an additional condition ($\chi^2 = .22$; $p > .05$). In the reference group, there were more boys (13.5%, $n = 127$) than girls (6.8%, $n = 60$; $\chi^2 = 22.46$, $p < .001$) in the high risk group.

Romantic Relationships (SPPA)

2×2×2 ANOVA: No interactions were found, and only one main effect, with boys reporting more positive perceptions of romantic appeal (EMM = 2.7, SE = .04) than girls (EMM = 2.4, SE = .05; F $(1,668) = 27.28$, $p < .001$; $\eta^2 = 0.039$). Cleft visibility did not have any impact on perceptions of romantic appeal (F $(1,668) = 2.49$, $p > .05$), nor did the presence of an additional condition (F $(1,668) = 3.34$, $p > .05$).

Norm group comparisons: Norms indicated that girls from the general population (M = 2.6; SD = .56) had more positive self-perceptions of romantic appeal than girls with a cleft (M = 2.4; SD = .63). However effect size was small (*d* = 0.27). There were no differences in means between norms for the boys (M = 2.7; SD = .51), and boys with a cleft (M = 2.7; SD = .67).

High risk group analysis: The cut-off point below the 10th percentile was calculated for the cleft group, with scores below 1.60 as indicative of high risk of negative perceptions of romantic appeal (10.5%, *n* = 71). There were more girls (17.1%, *n* = 43) in the high risk group than boys (6.5%, *n* = 25; χ^2 = 17.85, *p* < .001), and a somewhat higher frequency of adolescents with an additional condition (17.9%, *n* = 25 vs. 8.7%, *n* = 43; χ^2 = 9.71, *p* < .01). There were no differences related to cleft visibility (χ^2 = 1.01, *p* > .05).

Emotional Adjustment

Emotional adjustment was measured using the Depressive Symptoms Checklist, HSCL-7, and the Emotional Difficulties scale of the SDQ. Correlations between the measures used are provided in Table 2.

Depressive symptoms (HSCL-7)

2×2×2 ANOVA: As can be seen in Table 3, no interactions and only one main effect was found. Boys reported less depressive symptoms (EMM = 1.40, SE = .03) than girls (EMM = 1.72, SE = .04; F (1,690) = 45.59, *p* < .001; η^2 = 0.058). Cleft visibility did not account for any differences in depressive symptoms (F (1,690) = .02, *p* > .05), nor did the presence of an additional condition (F (1,690) = .17, *p* > .05).

Reference group comparisons: Compared to the reference group, adolescents with a cleft reported significantly less depressive symptoms. This was the case for both girls (Cleft group: M = 1.72, SD = .60; Reference group: M = 1.86, SD = .60; F (1,1154) = 11.64, p < .01; *d* =

0.23) and boys (Cleft group: M = 1.37, SD = .40; Reference group: M = 1.64, SD = .55; F (1,1368) = 84.83, p < .001; *d* = 0.56). Mean scores for girls from the reference group indicated depressive symptoms within the moderate range, while mean scores for the cleft group were within the normal range.

High risk group analysis: Scores above the cut-off for severe problems were categorised as high risk. There was a significantly higher frequency of adolescents from the reference group in the high risk group (23.3%, *n* = 426) than in the cleft group (11.8%, *n* = 82). While 20.5% (*n* = 55) of the girls with a cleft reported depressive symptoms within the severe range, this was the case for 6.3% (*n* = 27) of boys with a cleft (χ^2 = 32.04, *p* < .001), compared to 29.0% (*n* = 257) and 18% (*n* = 169) within the reference group (χ^2 = 30.99, *p* < .001). Cleft visibility was not associated with high risk for depressive symptoms (χ^2 = .84, *p* > .05), or the presence of an additional condition (χ^2 = .22, *p* > .05).

Emotional difficulties (SDQ)

2×2×2 ANOVA: No interactions were found in self- or parent reports. Self-reports revealed one main effect while there were two main effects indicated in parent reports (Table 3). Girls reported more emotional problems in self-reports (EMM = 3.9, SE = .19) and in parent reports (EMM = 2.4, SE = .20) than boys (Self-reports: EMM = 2.0, SE = .18; F (1,320) = 54.22, *p* < .001; Parents reports: EMM = 1.4, SE = .18; F(1,283) = 15.16, *p* < .001), with a large effect size in self-reports (η^2 = 0.128) and medium in parent-reports (η^2 = 0.042). In parent reports, the presence of an additional condition also had an impact on emotional problems (EMM = 1.3, SE = .16 and EMM = 2.5, SE = .21; F(1,283) = 18.81, *p* < .001; η^2 = 0.052).

Reference group comparisons: Compared to the reference group, girls with a cleft and no additional condition reported more emotional difficulties (M = 3.8, SD = 2.38) than girls from the reference group (M = 3.0, SD = 2.35; *d* = 0.34), while boys with a cleft and no additional

condition (M = 1.7, SD = 1.41) reported less emotional difficulties than boys from the reference group (M = 2.0, SD = 1.88; *d* = 0.18).

High risk group analysis: Cut-off points revealed 31 adolescents (9.4%) within the high risk group according to self-reports, while parents identified 29 adolescents in the high risk group (9.9%). According to self-reports, there were more girls (21.8%, *n* = 27) than boys (2.0%, *n* = 4; χ^2 = 35.57, *p* < .001) in the high risk group, as was the case in parent reports (17.4%, *n* = 19 and 5.5%, *n* = 10; χ^2 = 10.94, *p* < .01). There were no differences relating to cleft visibility in self-reports (χ^2 = 2.24, *p* > .05), while parents reported more problems in cases of a CP (16.1%, *n* = 14) and less with CLP (7.4%, *n* = 15; χ^2 = 7.22, *p* < .01). The presence of an additional condition was not associated with emotional risk according to self-reports (χ^2 = 4.05, *p* > .05), while parents reported a higher frequency of adolescents with emotional problems when the cleft was associated with an additional condition (6.0%, *n* = 12, 18.5%, *n* = 17; χ^2 = 11.0, *p* < .01).

<div align="center">

Satisfaction with appearance

</div>

Satisfaction with appearance was measured using the Physical Appearance scale of the SPPA and the Satisfaction with Appearance scale. Correlations between the measures used are provided in Table 2.

Physical appearance (SPPA)

2×2×2 ANOVA: No interactions and two main effects were found (Table 3). Girls reported more negative perceptions (EMM = 2.4, SE = .05) than boys (EMM = 3.0, SE = .05; F (1,702) = 73.86, *p* < .001). Effect size was medium to large (η^2 = 0.088). In addition, adolescents with a visible cleft reported more negative perceptions of physical appearance (EMM = 2.6, SE = .05) than adolescents with a non-visible cleft (EMM = 2.8, SE = .05; F (1,702) = 10.14, *p* <

.01). However, effect size was small ($\eta^2 = 0.012$). There were no differences relating to the presence of an associated condition ($F\ (1,702) = .01$, $p > .05$).

Reference group comparisons: Girls with a visible cleft ($M = 2.2$, $SD = .71$) reported similar self-perceptions of physical appearance as girls from the reference group ($M = 2.3$, $SD = .77$; $MD = -.04$, $SE = .07$, $p > .05$), while girls with CP reported significantly higher self-perceptions ($M = 2.6$, $SD = .82$) than girls with a visible cleft ($MD = .34$, $SE = .09$, $p < .01$; $d = 0.52$) and girls from the reference group ($MD = .30$, $SE = .07$, $p < .001$; $d = 0.14$). The differences between the groups of boys were minor (CP: $M = 3.0$, $SD = .58$; CLP: $M = 3.0$, $SD = .68$; Reference group: $M = 2.9$, $SD = .70$).

High risk group analysis: A total of 71 adolescents from the cleft sample (7.7%) were identified at high risk for dissatisfaction with appearance, compared to 10.2% ($n = 187$) of the adolescents in the reference group. As expected, there were more girls at high risk for negative perceptions (16.2%, $n = 59$) than boys (2.2%, $n = 12$; $\chi^2 = 60.98$, $p < .001$). The same distribution of gender was found in the reference group ($\chi^2 = 95.74$, $p < .001$). In spite of a significant main effect of cleft visibility on the ANOVA, adolescents with a visible cleft were not more frequently represented in the high risk group of negative perceptions of physical appearance than adolescents with a non-visible cleft ($\chi^2 = .17$, $p > .05$). There were no differences related to the presence of an additional condition ($\chi^2 = .85$; $p > .05$).

Satisfaction with Appearance (SWA)

2×2×2 ANOVA: No interactions and two main effects were found (Table 3). Girls were less satisfied with appearance (EMM = 6.7, $SE = .14$) than boys (EMM = 7.5, $SE = .13$; $F\ (1,558)$ = 19.34, $p < .001$; $\eta^2 = 0.030$). In addition, adolescents with a visible cleft reported less satisfaction with appearance (EMM = 6.3, $SE = .13$) than in cases of a non-visible cleft

(EMM = 7.9, SE = .15; (F (1,558) = 63.31, $p < .001$; $\eta^2 = 0.096$). There were no differences related to the presence of an associated condition (F (1,558) = .77, $p > .05$).

Lack of reference group: As far as the authors are aware, no norms exist for the SWA, and no studies have provided a reference group that would make comparisons with the current sample possible.

High risk group analysis: A risk group was identified using scores below the 10th percentile (M = 4.00). Fifty eight adolescents (10.3%) had scores below this cut-off point. More girls (14.7%, $n = 33$) than boys (7.4%, $n = 25$; $\chi^2 = 7.90$, $p < .01$) were found within the high risk group, and more adolescents with a visible cleft (13.3%, $n = 50$) than a non-visible cleft (3.3%, $n = 6$; $\chi^2 = 13.52$, $p < .001$). There was no effect related to the presence of an additional condition ($\chi^2 = .01$, $p > .05$).

<div align="center">

Risk groups across measures

</div>

In order to compare risk groups across measures in the cleft sample, five new variables were created[1]. The five new variables recorded the adolescents who had been identified as being at high risk of cognitive, behavioural, social and/or emotional problems, and/or at high risk for dissatisfaction with appearance, irrespective of which outcome measure had been used in the first instance. When categorising the adolescents in two groups (high risk versus borderline or normal range), a total of 53.1% of the adolescents ($n = 173$)[2] belonged to none of the high risk groups, 22.7% ($n = 74$) were at high risk in one group, 11.0% ($n = 36$) were at risk in two groups, 8.9% ($n = 29$) in three groups, 3.4% ($n = 11$) in four groups, and the last 0.9% ($n = 3$) were found to be at high risk in all five domains.

---

[1] Results related to the Global self-Worth (SPPA) and the Total score (SDQ) are based on the instruments' subscales, and were therefore not included in further analyses of high risk groups.

[2] Behavioural problems were only measured by the SDQ in the present study. As a consequence, sample size was smaller than for the other domains of adjustment. Hence, when calculating the number of participants across all risk groups, the total sample was reduced.

Additionally, adolescents reporting scores within the borderline range were identified and recorded. Hence, as an example, adolescents at high, borderline or no risk for dissatisfaction with appearance on the SWA and/or self-perceptions of physical appearance on the SPPA were recorded in the new variable named "Appearance-related risk". An overview of the frequency of adolescents with a cleft within the normal range, or in the borderline and high risk groups according to the five new variables is presented in Table 4.

In order to check for associations between domains of risk, the variables presented in Table 4 were used. Correlations between the five risk groups were calculated. All associations were significant. The strongest associations were found between appearance and emotional risk (r = .41, n = 690, p < .001) and between appearance and social risk (r = .37, n = 716, p < .001). The other correlations were, in order of strength of association: emotional and social risk (r = .34, n = 695, p < .001); cognitive and behavioural risk (r = .33, n = 332, p < .001); cognitive and emotional risk (r = .30, n = 694, p < .001); cognitive and social risk (r = .26, n = 721, p < .001); cognitive and appearance-related risk (r = .25, n = 715, p < .001); emotional and behavioural risk (r = .23, n = 332, p < .001); and behavioural and appearance-related risk (r = .22, n = 326, p < .001).

Discussion

To the authors' knowledge, the present study is the first to examine risk groups across cognitive, behavioural, emotional, social, and appearance-related domains of psychological adjustment within the same study, while also investigating patterns of co-variation between risk groups in order to explore whether risk can be understood to be general or domain-specific in adolescents with a cleft.

The prevalence of cognitive, behavioural, emotional, social, and appearance-related risk was significantly associated with gender on most measures, and in many cases was also related to

the presence of an additional condition. Cleft visibility, however, did not seem to affect adolescent adjustment negatively, except in relation to satisfaction with appearance. Overall, adolescents with a cleft had the same level of adjustment difficulties as same-aged adolescents from large national reference groups. Boys with a cleft had better adjustment scores, less peer problems and less depressive symptoms than boys from the reference group.

Half of the adolescents with a cleft (53.1%) were not at risk in any of the adjustment domains, one forth (22.7%) were at risk in one domain, while the last 25.2% were at risk in at least two domains of adjustment. Several significant associations were found, although on the whole the strength of these associations was low to moderate. The strongest associations were found between social and emotional risk, and between emotional risk and dissatisfaction with appearance. Overall, the results of the current study point towards risk and resilience as being domain-specific, rather than general, and to the fact that emotional, social, and appearance-related distress are normal during adolescence, whether there is a visible difference or not.

The results related to conduct difficulties must be interpreted with caution due to unacceptable internal reliability on this measure. The study provides insight into which of these measures may be useful in cleft clinics and in future research, discussed in more detail below.

Psychological adjustment: The role of an additional condition

Several outcome measures pointed to risk in cases where there was a condition affecting psychological adjustment in addition to the cleft. Although self-reports indicated no impact of an additional condition on adjustment, except in the case of cognitive function, adolescents with an additional condition were more at risk for general distress (Total score), emotional difficulties, peer problems, and (not unexpectedly) problems with attention, according to parent reports. In a similar study with children aged 10, a strong effect of the presence of an additional condition was demonstrated (Feragen and Stock, 2014). The wide range of

differing associated difficulties in children with a cleft raises the question of how to deal with co-morbid conditions in cleft research (Feragen, Stock, and Rumsey, in press). The results of the present study clearly support the need for early screening of children born with a cleft, in order to identify the children that may have associated difficulties, and who consequently may be at psychological and/or cognitive risk during childhood and adolescence.

Gender differences

Differences between boys and girls at age 16 were investigated within the cleft sample, and in comparison to the reference groups. A general finding was that girls reported more problems than boys on most measures, as is currently suggested in the general literature (Striegel-Moore and Franko, 2002; Van Roy et al., 2006). Specifically, girls with a cleft reported more emotional difficulties and poorer overall adjustment than boys with a cleft. In contrast to the general literature (Goodman, 1997; Galambos, 2004), and when comparing the cleft group to the reference group, boys in the cleft sample reported more positive perceptions of close relationships (SPPA) and less peer problems (SDQ) than girls in the cleft group. In addition, girls with cleft reported a similar quality of social experiences as the reference group. Many of the gender differences could be related to protective factors and therefore need to be discussed within the context of the other findings identified in the present study.

Risk and Protection

Social Experiences and Friendships

The findings related to social experiences and friendships were interesting, pointing to both risk and protective factors in the cleft sample. While visibility of cleft had a significant and negative impact on the girl's perceptions of peer problems on the SDQ, visibility of the cleft seemed to be associated with more positive perceptions of close friendships in boys with a

visible cleft than boys with a non-visible cleft, and boys from the reference group. These findings could point to protective factors in adolescent boys with a cleft, where the potentially negative social consequences of a visible difference are counteracted by close friendships, which in turn protects against emotional problems. The idea that a better quality of close friendships may act as a buffer against having difficulties with other peers has been suggested previously, both within the general literature (Berndt et al., 1999, Nangle et al., 2003) and within the CL/P literature (Baker et al., 2009; Feragen et al., 2010; Tiemens et al., 2013). In general, social support is known to be protective against psychological distress and to strengthen positive adjustment in individuals affected by a visible condition (Broder, 2001; Baker et al., 2009). These results are however in contrast to reports of social dysfunction (Nopoulos et al., 2005) and reduced quality of life in boys with a cleft (Mani et al., 2013). The present results of more positive social experiences could be associated with the fact that boys with a cleft reported less depressive symptoms than boys from the reference group. In addition, visibility of cleft did not affect adolescents' perceptions of social experiences. This could be indicative of protective factors, such as cognitive processes of appraisals (Bergen and Dalton, 2011; Tiemens et al., 2013), which mediate the relationship between objective visibility and social experiences, where a negative impact of visibility on social relationships is only perceived if the adolescent believe the social visibility to be problematic.

Romantic relationships

Boys with a cleft reported more positive self-perceptions of romantic appeal than girls with a cleft, as is reported in the general literature (Harter, 1988; Wichstrøm, 1998). More girls than boys with a cleft were found to be in the high risk group, and comparisons with norms indicated a slightly higher risk for negative self-perceptions of romantic appeal in girls with a cleft as compared to norms. Romantic intimacy is known to be important at this age (Collins,

2003), and may introduce new and additional challenges for adolescents with CL/P (Griffiths et al., 2012). However, calculations of effect size revealed that this difference in means was small. Although the findings within the cleft group could suggest that girls with a cleft are at risk in relation to perceptions of romantic appeal, perceptions of social acceptance and close friendships were the same as in the reference group, and could hence function as protective factors, moderating the impact of negative self-perceptions in other areas (Griffiths et al., 2012; Tiemens et al., 2013). The impact of perceptions of romantic appeal and the possible intervening factors could pose a question for future research.

Emotional difficulties

Overall, adolescents with a cleft reported less depressive symptoms than the reference group. Although girls with a cleft reported more problems than boys with a cleft, as is found in the general population, girls with a cleft reported scores within the normal range, while girls in the reference group reported scores within the 'moderate' range on the HSCL-7. On the SDQ, however, girls with a cleft reported more emotional problems than girls from the reference group. The discrepancy between these two measures could indicate that they capture somewhat different aspects of emotional adjustment. Nevertheless, the associations between the two measures were strong. Another explanation could be that comparisons were made with two different reference groups. Although internal reliability was shown to be acceptable for the emotional problems subscale of the SDQ, the SCL-7 appears to have better psychometric properties. As a consequence, results indicating that girls with a cleft had less depressive symptoms than same-aged adolescents appear to be supported.

In spite of the additional challenges associated with growing up with CL/P, adolescents with a cleft still appear to cope better than adolescents within the general population, as has been reported previously in children (Millar et al., 2013) and adults (Roberts and Mathias, 2013).

Such findings could be further indicative of processes of emotional resilience working within those born with CL/P. Exposure to challenges from a young age may result in psychological coping strategies in response to dealing with adversity (Baker et al., 2009), resulting in hardiness (Rutter, 2009).

Behavioural conduct

Both boys and girls in the cleft sample who did not have an additional condition reported less behavioural problems than boys and girls in the reference group. This effect was particularly large for boys with a cleft. These findings are in stark contrast to those which suggest that young people with a cleft, and boys in particular, are at higher risk of behavioural problems when compared to the general population (Richman and Millard, 1997). The differences in results between the present study and previous literature could be partly explained by the presence of a significant number of children with an additional condition in cleft samples (Feragen et al, in press), a factor that was controlled for in the present study.

Although internal consistency on self- and parent reports of behavioural conduct (SDQ) was deemed to be 'unacceptable' and findings must therefore be interpreted with caution, the items still appear to be measuring a large difference between adolescents with cleft and adolescents in the reference group. Further research with large comparable samples and other measures of behaviour may be necessary in order to assess the validity of the current findings.

Cognitive function

According to the current sample, adolescents with cleft were no more at risk of attention and/or hyperactivity problems than the reference group. In previous studies, a high risk of cognitive deficits in children with a cleft (Broder et al., 1998; Richman et al., 2005), as well as neurological abnormalities (Nopoulos et al., 2005; Nopoulos et al., 2007) has been

reported. In a recent study of psychological adjustment involving a large sample of 10-year-old children with CL/P, Feragen and colleagues identified that a high percentage of these deficits may be explained by the presence of a condition in addition to the cleft, rather than being a direct consequence of the cleft itself (Feragen et al., 2014). This again raises the need to be clear about who the participants are in a given study, and how representative that sample is. While the impact of poorer cognitive functioning on psychological adjustment should not be underestimated, it is possible that these reported deficits reflect this subgroup of children, rather than children with a cleft as a whole. However, the present results could also reflect the choice of measures. The SDQ only captures problems of attention and/or hyperactivity, rather than offering a more comprehensive assessment of cognitive function. In addition, scholastic competence was only measured via self-perceptions, in contrast to previous literature focusing on more objective measures of academic performance and/or parent and teacher reports (e.g. Persson et al, 2012).

Satisfaction with appearance

Despite the common belief that individuals with a cleft will be significantly less satisfied with their appearance than their peers without a cleft, the findings of the current study suggest that appearance is a concern for all young people at this stage of life, whether they have a visible difference or not. Although adolescents with a visible cleft were less satisfied with their appearance than adolescents with a non-visible cleft, all adolescents in the cleft group reported similar levels of appearance satisfaction as the reference group. These findings provide a useful example of the importance of including a reference or control group in studies such as these. If a reference group had not been available in the present study, the conclusion that girls with a visible cleft are at high risk of appearance-related distress would have been drawn.

These findings provide further support for other research in the area of visible difference, which suggests that subjective perceptions of appearance outweigh the objective severity of a condition (Moss et al., 2005; Appearance Research Collaboration, 2009; Mani et al., 2013). Individuals with a visible difference are likely to experience staring, comments and questions about their appearance, suggesting a social visibility that could be perceived as challenging by those affected (Moss, 2005; Tiemens et al., 2013). Future research is therefore needed in order to understand more about what could be the mediating factor(s) between cleft visibility and satisfaction with appearance.

Psychological adjustment

The existence of protective factors within the cleft group is summarised by the Total Score on the SDQ (self-reports) and the Global Self-Worth Scale on the SPPA. On both of these measures, girls with cleft reported similar overall psychological adjustment scores to the girls in the reference groups, while boys with a cleft reported less adjustment difficulties than boys in the reference groups. Internal consistency was acceptable/good for the Total Score and good/excellent for the SPPA, strengthening these overall findings of self-reported positive general adjustment in adolescents with a cleft.

Associations between risk groups

The strongest associations were found between social and emotional risk, as previously reported in the craniofacial literature (Roberts and Mathias, 2013), and between emotional risk and satisfaction with appearance. These associations may shed light on the previous finding that girls with a visible cleft report more negative social experiences such as peer harassment (Feragen and Borge, 2010). Within the present study, the highest frequency of adolescents at high or borderline risk was found within the domain of social experiences. Since the strongest association was found to be between social and emotional risk, results

could have indicated the presence of emotional problems. However, mean scores did not clearly indicate that girls with a cleft had more depressive symptoms or were more dissatisfied with appearance than the reference group. One possibility is that protective factors may be at work, such as positive coping strategies, close friendships and social support (Baker et al., 2009; Feragen et al., 2010; Berger and Dalton, 2011).

Although all of the associations between the different domains of risk were significant, most were weak to moderate. Overall, the findings of the present study therefore point to risk as being domain-specific, rather than general in adolescents with a cleft. Nonetheless, the fact that some associations exist suggests that interventions which foster improvements in one domain could also result in improvements in associated domains. For example, strengthening adolescents' perceptions of social competence may result in improvements relating to emotional resilience or satisfaction with appearance, as has been demonstrated previously (Kleve et al., 2002). Adjustment to a visible difference has been shown to fluctuate (Appearance Research Collaboration, 2009), suggesting that the impact of CL/P may become more or less salient during different periods of time. This could also help to explain why some of the associations between the different domains of risk were found to be weak. Ultimately, longitudinal studies are needed in order to understand how patterns of risk may vary from childhood to adolescence, and thus how support needs may change over time.

While one of the main findings of the current study was that adolescents with CL/P do not seem to experience psychological difficulties above that of their peers from the general population, it is clear that some adolescents are at risk of adjustment difficulties. More research is needed on subgroups of adolescents who might be at psychological risk in one or several domains of adjustment, in order to better understand why some adolescents with a visible difference do not cope with the challenges involved in their condition.

Psychometric properties

In general, the measures used in this study demonstrated good internal reliability. Nonetheless, the psychometric properties of some of the subscales of the SDQ were deemed to be unacceptable or questionable. While this indicates that some findings must therefore be interpreted with caution, the reports of reliability are in line with those reported previously (Goodman, 2001; Van Roy et al., 2008). In addition, relatively good correlations were observed between the majority of the different measures used. For example, although the peer problems scale of the SDQ was deemed to have unacceptable internal consistency, questioning the reliability of the finding that boys report less peer problems than the reference group; boys also reported more positive social experiences than the reference group on the SPPA, a measure which demonstrated good reliability.

For the SDQ and across the other measures, correlations between parent reports and self-reports were low to moderate. However, correlations were similar to those reported in previous studies of discrepancies between self- and parent reports (Van Roy et al., 2010), and point to the value of using a multidimensional approach to assessment. The SDQ is well-liked as it is easy and quick to administer, is widely available and free to use, and has been translated into several languages. Norms have been provided for many different countries, and reference groups are also available as a consequence of the number of studies using it. It is already used in some countries which have centralised cleft lip and palate treatment, which would make comparisons across countries possible and valuable in the future.

The SWA has been used in a number of cleft studies and was one of two measures in the present study which seemed to point to challenges related to cleft visibility. The measure is easy to administer and interpret, and demonstrated excellent internal reliability within the current sample. The SWA appears to be a useful measure, but again, unfortunately no published norms are available at present.

The CEQ has been used in other cleft studies, and therefore, comparisons across groups could be possible. Unfortunately, results are sometimes calculated in alternate ways across different studies, making meaningful comparisons more challenging. Although the psychometric properties of the CEQ were found to be good within the current sample of adolescents, it has been reported to have lower and more questionable reliability in younger children (Feragen et al., in press). In addition, the lack of norms makes the findings more difficult to interpret. Despite this, some cleft teams do find this measure clinically useful.

Both the SPPA and the HSCL-7 have been used in many studies and within different countries, and have been shown to have good psychometric properties. Although the SPPA has been used in some specific populations (e.g. chronic physical conditions; Aasland and Diseth, 1999), to the authors' knowledge, neither of the measures have been used widely within cleft research. It would be useful to see how the findings from the current study might compare to data collected in other countries, to explore whether the findings of this study could be related to cultural differences (Heiervang et al., 2008) or to having a centralised treatment system, or whether they are representative of the general cleft population.

Strengths and Limitations

The main strength of the current study was its large and representative sample of ten consecutive birth cohorts. The results are based on both self- and parent reports, with data being contributed by both mothers and fathers, which is rare in paediatric psychology research (Phares et al., 2005; Stock and Rumsey, in press). Unfortunately, parent reports were only available for the last four birth cohorts, and were not available for the reference groups. Another strength was that age was constant, thus controlling variables related to developmental differences. In addition, by running a 2×2×2 ANOVA instead of several *t*-tests when checking differences between variables, the chances of Type I error were kept at 5%.

Estimated marginal means that were calculated through the 2×2×2 ANOVA were also adjusted for the other variables in the model and hence provide more correct estimations of a variable's effect on an outcome.

Several limitations have to be considered. The main limitation was the lack of a control group. However, this was offset by access to large national reference groups, which allowed for comparisons across groups to be made. In addition, by discussing the limitations of each measure in relation to the results produced, any limitations in relation to the measures, such as questionable internal reliability, were partially counteracted. Third, the reference group data were collected in 2002, while the cleft sample was assessed 2004-2013. Hence, potential changes in societal attitudes and/or adolescent health from 2002 and onwards were not represented in the reference group. However, the impact of this limitation was considered to be small, since only a decade separated the two groups. Another limitation could be the lack of data for the adolescents with severe developmental problems who did not go through the routine assessment. Their presence in the sample would probably have impacted on the mean scores of most variables and needs to be acknowledged. However, since the present study controlled for the effect of an additional condition, their presence in the sample would only have affected this subgroup of adolescents. Fifth, adjustment to a visible difference involves a combination of psychological and societal factors, such as individual characteristics and cognitive processes (such as personality, attribution style or coping strategies), family factors and social support, in addition to socio-cultural factors. These variables were not included in the present study and future research should aim to include such information. Last, the present study could be criticised for including measures with slightly different foci within the same domains of risk (e.g. scholastic competence and attention/hyperactivity problems). While this needs to be explored further within future studies, associations between the measures were reasonably good, indicating that different measures were accessing some of the same aspects

of adjustment. Another example could be related to the finding that half of the adolescents with a cleft were at social risk, indicating that adolescents with a visible difference might be at high risk of social difficulties. While this could be an accurate finding, it may also reflect the use of five different measures, all capturing slightly different aspects of social experiences. By using several measures in this way, risk may have accumulated, increasing the number of children identified as being at risk when summarising the findings within the social domain.

Summary and conclusions

The objectives of the present paper were to investigate whether there were associations between different domains of risk at age 16 and to explore the usefulness of measures of psychological adjustment across a range of domains. Approximately half of the adolescents were not at risk on any adjustment measures, while a forth were at risk in two or more domains of adjustment. However, findings were comparable to those derived from large national reference groups. Gender was the most prominent mediating factor in the current study, as has been reported in studies with adolescents in general. Adolescents with a condition in addition to the cleft were also at higher risk on a number of measures, indicating the importance of early screening and assessment, in order to diagnose possible associated conditions and offer adapted and appropriate treatment and care. Associations were found between social and emotional risk, and between emotional risk and appearance-related distress. Overall, risk appeared to be domain-specific, rather than general. A number of possible protective factors were also discussed. Finally, this study has examined a number of measures pertaining to psychological adjustment at age 16 in relation to clinical relevance and psychometric value.

Reference List

Aasland A, Diseth TH. Can the Harter Self-Perception Profile for Adolescents (SPPA) be used as an indicator of psychosocial oucome in adolescents with chronic physical disorders? *Eur Child Adolesc Psychiatry*. 1999;8:78-85.

Appearance Research Collaboration (ARC). *Identifying factors and processes contributing to successful adjustment to disfiguring conditions*. Bristol: Centre for Appearance Research, University of the West of England; 2009. Final report.

Baker SR, Owens J, Stern M, Willmot D. Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *Cleft Palate Craniofac J*. 2009;46:229–236.

Berger ZE, Dalton LJ. Coping with a cleft: Psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *Cleft Palate Craniofac J*. 2009; 46:435–443.

Berger ZE, Dalton LJ. Coping with a cleft II: Factors associated with psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *Cleft Palate Craniofac J*. 2011;48:82–90.

Berndt TJ, Hawkins JA, Jiao Z. Influences of friends and friendships on adjustment to junior high school. *Merrill-Palmer Quarterly*. 1999;45:13–41.

Broder HL. Using psychological assessment and therapeutic strategies to enhance well-being. *Cleft Palate Craniofac J*. 2001;38:248-254.

Broder, HL, Richman LC, Matheson PB. Learning disability, school achievement and grade retention among children with cleft: A two-centre study. *Cleft Palate Craniofac J*. 1998;35:127-131.

Cohen J. Statistical power analysis for the behavioral sciences (2nd ed). Lawrence Erlbaum Associates.

Collett BR, Speltz ML. A developmental approach to mental health for children and adolescents with orofacial clefts. *Ortho Craniofac Res.* 2007; 10:138-148.

Collins WA. More than myth: The developmental significance of romantic relationships during adolescence. *J Res Adolesc.* 2003;13:1-24.

Dekker MC, Ferdinand RF, Van Lang NDJ, Bongers IL, Van Der Ende J, Verhulst FC. Developmental trajectories of depressive symptoms from early childhood to late adolescence: gender differences and adult outcome. *J Child Psychol Psychiatry*. 2007; 48:657-666.

Emerson M, Spencer-Bowdag S, Bates A. Relationships between self-esteem, social experiences and satisfaction with appearance: Standardisation and construct validation of two cleft audit measures. Presented at *The Craniofacial Society of Great Britain and Ireland's Annual Scientific Conference, ''Setting a Positive Agenda in Cleft and Craniofacial Care''*; April, 2004; Bath, United Kingdom.

Feragen KJB, Borge AIH. Peer harassment and satisfaction with appearance in children with and without a facial difference. *Body Image*. 2010;7:97-105.

Feragen KJB, Kvalem IL, Rumsey N, Borge AIH. Adolescents with and without a facial difference: The role of friendships and social acceptance in perceptions of appearance and emotional resilience. *Body Image.* 2010; 7:271-279.

Feragen KJB, Stock NM. When there is more than a cleft: Psychosocial adjustment in children with an associated condition. *Cleft Palate Craniofac J.* 2014; 51:5-14.

Feragen KJB, Stock NM, Rumsey N. Toward a reconsideration of inclusion and exclusion criteria in cleft lip and palate: Implications for psychological research. *Cleft Palate Craniofac J.* Epub ahead of print; doi: http://dx.doi.org/10.1597/12-326.

Galambos NL. Gender and gender role development in adolescence. In: RM Lerner and L Steinberg, eds. *Handbook of adolescent psychology*. New Jersey: John Wiley & Sons, Inc; 2004:233-262.

Goodman R. The Strengths and Difficulties Questionnaire: A research note. *J Child Psychol Psychiatry*. 1997;38:581-586.

Goodman R. Psychometric properties of the strengths and difficulties questionnaire. *J Am Acad Child Adolesc Psychiatry*. 2001;40:1337-1345.

Griffiths C, Williamson H, Rumsey N. The romantic experiences of adolescents with a visible difference: Exploring concerns, protective factors and support needs. *J Health Psychol.* 2012;17:1053-1064.

Harter S. *Manual for the Adolescent Self-Perception Profile.* University of Denver: 1988.

Heiervang E, Goodman A, Goodman R. The Nordic advantage in child mental health: Separating health differences from reporting style in a cross-cultural comparison of psychopathology. *J Child Psychol Psychiatry*. 2008;49:678-685.

Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. *Eur J Orthod.* 2005; 27:274-285.

Kandell DB, Davies M. Epidemiology of depressive mood in adolescents: An empirical study. *Archives of General Psychiatry*. 1982;39:1205-1212.

Kleve L, Rumsey N, Wyn-Williams M, White P. The effectiveness of cognitive-behavioural interventions provided at outlook: A disfigurement support unit. *J Eval Clin Practice.* 2002;8:387–395.

Kumpfer KL. Factors and processes contributing to resilience: The resilience framework. In: Glantz MD, Johnson JL (Eds), *Resilience and development: Positive life adaptations.* New York: Klumer Academic/Plenum Publishers. 1999:179-224.

Locker D, Jakovic A, Tompson B. Health-related quality of life of children aged 11 to 14 years with orofacial conditions. *Cleft Palate Craniofac J.* 2005;42:260-266.

Luthar SS. Resilience in development: A synthesis of research across five decades. In: D Cicchetti, DJ Cohen (Eds.), *Developmental Psychopathology: Risk, disorder, and adaptation* (2nd edition). New York: Wiley; 2006:739-795.

Mani M, Reiser E, Andlin-Sobocki A, Skoog V, Holmström M. Factors related to quality of life and satisfaction with nasal appearance in patients treated for unilateral cleft lip and palate. *Cleft Palate Craniofac J.* 2013;50:432-439.

Masten AS. Ordinary magic: Resilience processes in development. *American Psychologist.* 2001; 56:227-238.

Milerad J, Larson O, Hagberg C, Ideberg M. Associated malformations in infants with cleft lip and palate: A prospective population-based study. *Pediatrics.* 1997;100:180-186.

Millar K, Bell A, Bowman A, Brown D, Lo T-W, Siebert P, Simmons D, Ayoub A. Psychological status as a function of residual scarring and facial asymmetry after surgical repair of cleft lip and palate. *Cleft Palate Craniofac J.* 2013;50:150-157.

Millard T, Richman LC. Different cleft conditions facial appearance and speech: relation to psychological variables. *Cleft Palate Craniofac J.* 2001;38:68–75.

Moss TP. The relationships between objective and subjective ratings of disfigurement severity and psychological adjustment. *Body Image.* 2005;2:151-159.

Nangle DW, Erdley CA, Newman JE, Mason CA, Carpenter EM. Popularity, friendship quantity and friendship quality: Interactive influences on children's loneliness and depression. *J Clin Child Adolesc Psychol.* 2003;32:546-555.

Nolen-Hoeksema S, Girgus JS. The emergence of gender differences in depression during adolescence. *Psychol Bull.* 1994;115:424-443.

Nopoulos P, Choe I, Berg S, Van Demark D, Canady J, Richman L. Ventral frontal cortex morphology in adult males with isolated orofacial clefts: relationship to abnormalities in social function. *Cleft Palate Craniofac J.* 2005;42:138-44.

Nopoulos P, Langbehn DR, Canady J, Magnotta V, Richman LC. Abnormal brain structure in children with isolated clefts of the lip or palate. *Arch Pediatr Adolesc Med.* 2007;161:753-758.

Nurmi J-E. Socialization and self-development. Channeling, selection, adjustment, and reflection. In: Lerner RM, Steinberg L, eds. Handbook of adolescent psychology, Hoboken, New Jersey: John Wiley and Sons, 2004:85-124.Paxton SJ, Eisenberg ME,

Paxton SJ, Neumark-Sztainer D, Hannan PJ, Eisenberg ME. Body dissatisfaction prospectively predicts depressive mood and low self-esteem in adolescent girls and boys. *J Clin Child Adolesc Psychol.* 2006;35:539-549.

Persson M, Becker M, Svensson H. Academic achievement in individuals with clefts: A population-based register study. *Cleft Palate Craniofac J.* 2012;49:153-159.

Pertschuk MJ, Whitaker LA. Social and psychological effects of craniofacial deformity and surgical reconstruction. *Clin Plast Surg.* 1982;9:297–306.

Phares V, Lopez E, Fields S, Kamboukos D, Duhig AM. Are fathers involved in pediatric psychology research and treatment? *J Pediatr Psychol.* 2005;30:631-643.

Pope AW, Snyder HT. Psychosocial adjustment in children and adolescents with a craniofacial anomaly: Age and sex patterns. *Cleft Palate Craniofac J.* 2005; 42:349-54.

Richman LC, Millard T. Cleft lip and palate: longitudinal behavior and relationships of cleft conditions to behavior and achievement. *J Pediatr Psychol*. 1997;22:487–494.

Richman LC, Wilgenbusch T, Hall T. Spontaneous verbal labeling: visual memory and reading ability in children with cleft. *Cleft Palate Craniofac J.* 2005;42:565-569.

Roberts RM, Mathias JL. Predictors of mental health in adults with congenital craniofacial conditions attending the Australian Craniofacial Unit. *Cleft Palate Craniofac J.* 2013;50:414-423.

Roberts RM, Mathias JL, Wheaton P. Cognitive functioning in children and adults with nonsyndromal cleft lip and/or palate: A meta-analysis. *J Pediatr Psychol.* 2012;37:786-797.

Rognerud M, Strand BH, Dalgard OS. Psykisk helse i Helse- og levekårsundersøkelsen i 1998. 1. Sosioøkonomiske forskjeller i psykisk helse og livsstil *[Mental health in Norway 1998. 1. Socioeconomic differences in mental health and lifestyle]. Norsk Epidemiologi.* 2002;12:239-248.

Rumsey N, Harcourt D. *The Psychology of Appearance*. Berkshire: Open University Press. 2005.

Rumsey N, Stock NM. Living with a cleft: Psychological challenges, support and intervention. In: S. Berkowitz, ed. *Cleft Lip and Palate: Diagnosis and Management (third edition)*. Springer-Verlag Berlin Heidelberg; 2013:907-17.

Rutter M. Understanding and testing risk mechanisms for mental disorders. *J Child Psychol Psychiatry*. 2009;50:44–52

Rønning JA, Handegaard BH, Sourander A, Mørch WT. The strengths and difficulties self-report questionnaire as a screening instrument in Norwegian community samples. *Eur Child Adol Psych.* 2004;13:73-82.

Speltz ML, Endriga MC, Hill S, Maris CL, Jones K, Omnell ML. Cognitive and psychomotor development of infants with orofacial clefts. *J Pediatr Psychol.* 2000;25:185-190.

Striegel-Moore R, Franko D. Body image issues among girls and women. In: TF Cash and T Pruzinsky (Eds.), *Body Image: A handbook of theory, research and clinical practice*. New York: Guilford Press. 2002.

Stock NM, Rumsey N. Parenting a child with a cleft: The father's perspective. *Cleft Palate Craniofac J*. Epub ahead of print; doi: http://dx.doi.org/10.1597/13-035.

Swanenburg dV, Beemer FA, Mellenbergh GJ, Wolters WH, Heineman-de Boer JA. An investigation of the relationship between associated congenital malformations and the mental and psychomotor development of children with clefts. *Cleft Palate Craniofac J.*, 2003; 40:297-303.

Tambs K, Moum T. How well can a few questionnaire items indicate anxiety and depression? *Acta Psychiatrica Scandinavica.* 1993;87:364-367.

Tiemens K, Nicholas D, Forrest CR. Living with difference: experience of adolescent girls with cleft lip and palate. *Cleft Palate Craniofac J.*, 2013;50:e27-e34.

Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Self-reported strengths and difficulties in a large Norwegian population 10-19 years. Age and gender specific results of the extended SDQ-questionnaire. *Eur Child Adol Psychiatry.* 2006;15:189-198.

Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Understanding discrepancies in parent-child reporting of emotional and behavioural problems: Effects of relational and socio-demographic factors. *BMC Psychiatry*. 2010;10:56.

Van Roy B, Veenstra M, Clench-Aas J. Construct validity of the five-factor Strengths and Difficulties Questionnaire (SDQ) in pre-, early, and late adolescence. *J Child Psychol Psychiatry.* 2008;49:1304-1312.

Wichstrøm L. Harter's Self-Perception Profile for Adolescents: Reliability, validity and evaluation of the question format. *J Pers Assess*. 1995;65:100-116.

Wichstrøm L. Self-concept development during adolescence. Do American truths hold for Norwegians? In: E. Skoe & A.v.d. Lippe, Eds. *Personality development in adolescence. A cross national and life span perspective.* Routledge, London; 1998:98-122.