Risk and protective factors at age 10: Psychological adjustment in children with a cleft lip and/or palate

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Abstract

Objective: Explore psychological functioning in children with a cleft at age 10 from a broad perspective, including cognitive, emotional, behavioural, appearance-related and social adjustment. High risk groups were identified within each area of adjustment, in order to investigate whether vulnerable children were found across domains, or whether risk was limited to specific areas of adjustment.

Methods: Retrospective chart-review from psychological assessments at age 10 (n=845). The effects of gender, cleft visibility and the presence of an additional condition were investigated. Results were compared to large national samples.

Measures: Personality Inventory for Children, Child Experience Questionnaire, Strengths and Difficulties Questionnaire, Satisfaction with Appearance scale.

Results: The factor affecting psychological adjustment on most domains was the presence of an associated condition in addition to the cleft. As expected, no support was found for cleft visibility as a risk factor, while there were some gender differences related to emotional difficulties and attention. Correlation analyses of risk groups pointed to an association between social experiences and emotional adjustment and between social and behavioural adjustment, while dissatisfaction with appearance was not related to any other domains of risk at age 10.

Conclusions: The results point to the importance of early screening and assessment of children born with a cleft, in order to identify possible associated conditions and offer adapted and appropriate treatment and care. Future research should investigate how protective factors could counteract potential risk in children with a cleft.

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

Psychological research on cleft lip and/or palate (CL/P) currently provides an inconsistent picture of how individuals adjust to this condition. Some studies point to children who may be at risk within particular areas of psychological functioning, such as dissatisfaction with facial appearance, cognitive performance, behavioural difficulties and social and emotional experiences (see review papers such as Turner et al., 1998; Thompson and Kent, 2001; Hunt et al., 2005). However, more recent studies have also reported a number of positive outcomes. These findings highlight a number of possible protective factors and illustrate the potential for the development of resilience within children and adolescents with a cleft (Baker et al., 2009; Berger and Dalton, 2009; Feragen et al., 2009; Kramer et al., 2009). While mixed findings almost certainly highlight the notion of adjustment as a multifaceted and complex process, they are also a likely consequence of a wide variation in concepts and instruments.

Although studies often aim to investigate the same areas of psychological adjustment, there is a clear discrepancy in the measures which are used (Klassen et al., 2012; Rumsey and Stock, 2013), complicating comparisons between studies. In addition to the need for comparisons, there is a need to agree upon measures which would help researchers to discriminate clearly between those children with CL/P who cope well and those who may be at risk. One additional consideration in regard to choosing instruments is whether to use generic measures of psychological wellbeing or more condition-specific measures. While generic measures provide universal information that can be compared to reference groups and control groups, specific measures may be more sensitive to the aspects and challenges associated with a particular condition (Roberts and Shute, 2011). Although there is a probability that a combination of both types of measures would be most helpful, clear guidance is not available due to the current lack of consistency within research findings. Agreeing on measures is a cumbersome process, involving different possibilities and restrictions in clinical settings, as

well as cultural differences, to name a few. This dialogue is therefore on-going among cleft clinicians and researchers.

In addition to the ability to identify children at risk, a fundamental background factor of any measure should be its psychometric strengths and weaknesses. In order to determine a measure's psychometric value, large samples are needed. Only a minority of studies are able to include a dataset that is comprehensive enough to fully evaluate psychometric merit. In addition, very few papers discuss their findings within the context of the psychometric properties of the measures they have used. This insight may be particularly interesting and necessary when a measure or a subscale has been shown to have questionable validity and/or reliability in a previous study. The psychometric qualities of the measures used may be an additional contributory factor to the acquisition of mixed findings in the field.

A second point of discussion relates to the actual process of adjustment. Discrepancies in research findings may be partly reflective of different domains of risk and resilience working within the same individual. The fact that children may be at risk in some domains while demonstrating good adjustment in other areas has been established within the general resilience literature (see Luthar, 2006; Masten, 2001). Although psychological research within the field of CL/P has not yet specifically addressed this question, studies have attempted to look at associations between different areas of adjustment (e.g. Berger and Dalton, 2011). Unfortunately, studies often only investigate adjustment across one or two domains. This makes it difficult to know whether those children who are at risk of, for example, appearance dissatisfaction or social difficulties, are also at risk in other domains of psychological health. Looking at adjustment across a range of different domains would make it possible to compare risk groups across measures, and to investigate whether co-variations between risk groups might exist, or whether a lack of associations between areas of risk could be an indicator of protective factors. Information about specific or potential risk and

protective factors might assist primary care providers and cleft teams in targeting those children and families who may need more intensive care, while at the same time being able to capitalize on strengths and resilience factors, hence utilising limited resources more efficiently. To date, little research has aimed to explore both risk and protective factors within the same study.

A number of additional factors have produced interesting findings within the adjustment literature and therefore warrant further investigation. CL/P is associated with a relatively high prevalence of additional conditions which are known to impact on psychological functioning (Broder, 1997; Baker et al., 2009; Feragen et al., in press), such as developmental difficulties, or a range of milder conditions, such as attention deficit and/or hyperactivity disorder (AD/HD) or dyslexia. Recent research has indicated that this group of children may be at increased psychological risk (Feragen and Stock, 2014). Therefore, in order to help differentiate between the consequences of being born with CL/P, and the consequences of having an associated difficulty, additional conditions need to be identified and categorised accordingly, and accounted for in a study's methodology. At present, virtually no studies have taken this potentially confounding variable into account in their methodology (Feragen et al., in press).

Research within the general literature has also highlighted a number of potential gender differences among children and adolescents. For example, girls often report more emotional difficulties and higher levels of appearance dissatisfaction, while boys report more conduct and peer problems (Van Roy et al., 2006, 2010). In the cleft literature, conflicting results have been reported (Berger and Dalton, 2011; Klassen et al., 2012). Since a visible cleft is significantly more frequent in boys, studies focusing on cleft types need to take this factor into account. In addition, age may be a confounding factor, since studies often use samples of children who are at different developmental stages.

The question of whether the visibility of a cleft impact on adjustment has created much debate within the field. Although a number of studies have indicated that an individual's subjective feelings about appearance outweigh the objective severity of a visible difference (Appearance Research Collaboration, 2009; Feragen et al., 2010; Moss, 2005), many papers continue to investigate visibility as a key variable (Broder et al., 1994; Millard and Richman, 2001; Berger and Dalton, 2009; Mani et al., 2013). In addition, some general differences between cleft types have been observed. For example, children with palatal involvement are often shown to have greater or differing cognitive difficulties than their peers with other cleft types, and when compared to matched comparison groups (Speltz et al., 2000; Christensen and Mortensen, 2002; Roberts et al., 2012). Some studies have also suggested differences between bilateral and unilateral clefts (Millard et al., 2001). However, with respect to psychological adjustment, most reported differences involving cleft types are related to cleft palate vs. cleft lip and palate (for a review, see Hunt et al. 2005). From a psychological perspective, a classification of cleft types as visible vs. non-visible therefore seems adequate.

In order to explore whether risk and resilience may co-vary within the same individual, a comprehensive perspective on adjustment is necessary. Further, the impact of gender, visibility of cleft, and the presence of an associated condition might vary, depending of the domain of psychological adjustment under study. Several recent review papers and book chapters provide an extensive overview of domains of psychological adjustment that have been shown to be important in cleft research and are considered central during childhood (Thompson and Kent, 2001; Hunt et al., 2005; Feragen, 2012; Klassen et al., 2012; Richman et al., 2012; Rumsey and Stock, 2013). Domains related to outcome (in contrast to predisposing and intervening factors such as personality, coping strategies, or sociocultural factors) were found to include general adjustment, self-concept and self-esteem, satisfaction

with speech and appearance, behaviour, social functioning and experiences, emotional distress, quality of life, and school-related/cognitive functioning.

The aims of the present study were: First, to explore adjustment across a wide range of domains. Among all identified domains that were mentioned above, measures of quality of life and self-concept were not available in the present study. However, all other aspects of psychological adjustment were represented and categorised into five main domains: cognitive, behavioural, emotional and social functioning, and satisfaction with appearance. The effects of gender, cleft visibility and the presence of an additional condition were evaluated, in addition to possible interactions for each of the five domains. Second, 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. Third, to present and discuss psychometric properties in relation to each outcome variable.

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.

Method

Setting

The present study was based on a retrospective clinical audit review of case records of 10-year-old children with cleft lip and/or palate, from a centralised treatment setting. Patient confidentiality was preserved, and the Regional Committee for Medical Research Ethics granted ethical approval for the study. The team's clinical psychologist conducted the psychological assessment. If needed, the child could be helped to complete the questionnaires. All measures used in the present study were administered as part of routine care. The assessment also includes a dialogue with the child's parent(s).

Participants

Children

All children (*n* = 845) who attended the routine 10-year-old follow-up from August 2002 to December 2013 were eligible for inclusion in the study, hence 11 and a half consecutive birth cohorts. No participants were excluded from the study. However, due to severe developmental problems, some children (*n* = 51) were not able to attend the routine assessments and most outcome measures are missing.

In the cleft sample, 336 children were female and 509 were male. Children's cleft type included cleft lip and palate, CLP (*n* = 368), cleft lip or cleft lip alveolus, CLA (*n* = 120)[1], cleft palate, CP (*n* = 275) or submucous cleft palate, SMCP (*n*= 59). Information about the child's cleft type was missing for three children. For the purpose of the statistical analyses, the children were categorized into two groups: children with visible clefts (CLP and CLA, *n* = 488) and children with non-visible clefts (CP/SMCP, *n* = 354). Among the girls, 51.8% had a non-visible cleft and 48.2% had a visible cleft. Among the boys, 31.4% had a non-visible cleft and 68.6% had a visible cleft. Some of the children were of non-Caucasian origin (*n* = 86/812, 10.6%), some of them adopted (*n* = 55/798; 6.9% of the total sample).

Parents

A total of 722 parents participated in the study by completing the (Nationality) version of the Parent Questionnaire (developed by the Psychology Special Interest Group of the Craniofacial Society of Great Britain and Ireland, CFSGBI), and (from 2010 onwards) also the Strengths and Difficulties Questionnaire (SDQ). A total of 153 did not report their relation to the child. Among the 569 who did, 30% (*n* = 168) were fathers, 51% were mothers (*n* = 288), or both

[1] Children with CL/CLA were, until April 2007, not offered a psychological follow-up at age 10. Thus, children with CL/CLA are missing in the birth cohorts from 1992 to 1997.

parents together (*n* = 102, 18%). The eleven respondents (2%) who were not the child's parents included siblings, grandparents and foster parents.

Additional conditions

Information about additional conditions was found in the child's case records, discussed during the 10-year-old assessment, and/or was given by the child's parents. A total of 278 children (33.3%) had one or several additional conditions, such as developmental delay (13.4%; *n* = 114), learning difficulties (7.3%; *n* = 62), dyslexia (5.5%; *n* = 47), autism spectrum disorders (1.9%; *n* = 16) and AD/HD (8.0%; *n* = 68). Furthermore, some children had a diagnosed syndrome (9.3%; *n* = 79/847), such as 22Q11.2, Treacher Collins, Goldenhar and Sticklers, with or without associated psychological and/or cognitive difficulties. While 135 of the children had one extra diagnosed condition in addition to the cleft (16.1% of the total sample; 48.4% of the children with an additional condition), 79 of the children had two additional diagnoses (9.4%; 28.3%), while the remaining 65 had three or more conditions in addition to the cleft (7.7%; 23.3%).

Measures

Personality Inventory for Children (PIC)

The PIC (Wirt et al., 1984) is a multidimensional personality inventory consisting of 280 true-false items. It provides good coverage of psychosocial adjustment through various behavioural, cognitive, emotional and interpersonal domains, using the child's mother as the informant. The PIC provides an empirical classification based on 12 clinical scales, placing a T-value within normal limits, or within the category of mild, moderate or severe problems. The clinical scales that were used were those known to be clinically useful and relevant for the five domains of adjustment that were the focus of the present study: the general Adjustment scale, Intellectual Screening, Withdrawal, Hyperactivity, Depression and Anxiety scales. The Intellectual Screening scale has been reported to correlate -.55 with the Full Scale

IQ on the Wechsler scales (Wirt et al., 1984). A Norwegian version of the instrument was used (Troland, 1988). Internal consistency (α = .59-.86; M = .74), test-retest reliability (r = .46-.94; M = .86), and validity have been extensively evaluated and found to be satisfactory (Wirt et al., 1984).

Child Experience Questionnaire (CEQ)

The CEQ (Pertschuk & Whitaker, 1982) reflects the child's self-reporting of social experiences on a 5-point Likert scale. The questions in the scale relate to topics such as relationships with friends ("I play with friends at school"), social isolation ("I try to hide from people"), and involvement in new experiences ("I meet new people"). Both positively and negatively worded items are included, 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 et al., 2004).

Strength and Difficulties Questionnaire (SDQ)

The SDQ (Goodman, 1997) is a screening tool for behavioural difficulties and strengths in children. The SDQ was completed by both the parent(s) and the child, since both informants are important to minimise the false negatives (Van Roy et al., 2010). The SDQ includes five subscales measuring emotional distress, conduct problems, hyperactivity/attention difficulties, peer relationship problems and pro-social behaviour. Each subscale includes five items that are positively or negatively worded. Each item is scored "not true", "somewhat true" or "certainly true" (0-2). The first four subscales are summarized into the Total difficulties score (including 20 items in total, with a total score ranging from 0-40). Internal consistency has been reported to range from .44 to .61 (M = .54) in same-aged children on self-reports, and from .50 to .76 (M = .62) on parent/proxy reports (Van Roy et al., 2010). Cut-off points for identifying children at risk are recommended to be set at the 90th percentile. The SDQ has

257 been extensively validated, and cut-off scores presented by Goodman (www.sdq.info) have
258 been slightly adjusted to a (Nationality) population and are the ones used as a reference in the
259 present study (Van Roy et al., 2006).

260 *Satisfaction With Appearance (SWA)*

261 The SWA (developed by the Psychology Special Interest Group of the CFSGBI) reflects
262 satisfaction with cleft-related and non-cleft-related parts of the face, speech, overall
263 appearance and the perceived visibility of the cleft. Each rating is made on an interval scale
264 of 0 to 10 where a score of 10 indicates very high levels of satisfaction with appearance. The
265 mean total score of a 12 item version of the scale was used in the present study (Range 0-10).
266 The SWA has been reported to possess satisfactory internal consistency and a coherent factor
267 structure (Emerson et al., 2004).

268 Statistical Analyses

269 SPSS 21 was employed for the statistical analyses. The first part of the results investigates
270 the outcome variables according to the study's aims, and the identification of high risk
271 groups. In order to enhance readability, the results are presented in the following order for
272 each outcome variable:

273 i. A 2 × 2× 2 ANOVA exploring the main effects and potential interactions of
274 gender, cleft visibility and the presence of an additional condition on the
275 outcome variable. The ANOVA provides adjusted effects of means (EMM)
276 and standard errors (SE), and avoids an accumulation of Type I errors as
277 would be the case with successive *t*-tests. In order to assess the magnitude of
278 the findings, Eta square effect sizes (η^2) were calculated. Cohen's guidelines
279 (1988) were used to interpret η^2: small effect: 0.01; medium effect: 0.059;
280 large effect: 0.138. Effect sizes were only calculated in cases of statistical

significance. Statistically non-significant findings are only reported in the table.

ii. Comparisons between the cleft sample and reference groups/norms and/or clinical cut-off scores are given. Reference groups for the SDQ were large national same-aged and non-cleft samples (Self-reports: Van Roy et al., 2006; Parent reports: Van Roy et al., 2010), which were compared to children with a cleft and no additional condition. Independent sample *t*-tests provided Mean scores (M) and Standard deviations (SD) which could be directly compared with scores from the reference group. Calculations of effect size were performed using Cohen's *d* in cases of significant differences (Cohen, 1988; 0.2 = small, 0.5 = medium, and 0.8 = large effect).

iii. Identification of a high risk group according to norms (PIC: clinical cut-off scores indicating moderate or severe problems) or according to scores below the 10^{th} percentile (SWA and CEQ) or above the 90^{th} percentile (SDQ). Cut-off scores from large national samples were used for the SDQ. 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.

In the second part of the results, five new variables were created based on the identification of the risk groups within each measure, classifying risk according to the different domains of adjustment (cognitive, behavioural, social, emotional, and appearance-related). In addition to the identified high risk group presented in the first part of the results, borderline cases were also identified. The SDQ provides cut-off scores within the borderline range, while cut-off scores identifying children with mild

problems were used for subscales on the PIC. Two measures do not provide norms (CEQ and SWA). For these two measures, scores between the 10th and the 25th percentile were categorized as borderline. Hence, the five new variables identified children scoring within the normal, borderline, or high risk range within each domain of adjustment. In order to investigate a potential co-variation between the risk groups, Pearson's correlation coefficients were used.

In the third and last part of the results, concurrent validity was explored by calculating Pearson's correlation between subscales that measure similar dimensions, across measures and across informants (children and parents). In addition, calculations of internal reliability for all subscales were calculated and presented.

Results

General Adjustment

General adjustment was measured through the Adjustment scale of the PIC and the Total difficulties score of the SDQ (self- and parent reports).

Adjustment (PIC)

2×2×2 ANOVA: There were no interactions, while two main effects were found, related to cleft visibility and the presence of an additional condition (Table 1). Children with a CP (with and without an additional condition) had significantly less adjustment problems (EMM = 55.9, SE = .84) than the total sample of children with CLP (EMM = 58.4, SE = .86; F (1,435) = 4.12, $p < .05$). However, calculations of effect size showed that this effect was small ($\eta^2 = 0.007$). A main effect was also found between children with a cleft only (EMM = 50.6, SE = .64) and children with a cleft and an additional condition (Cleft +: EMM = 63.7, SE = 1.02; F (1,435) = 116.95, $p < .001$), with a very large effect size ($\eta^2 = 0.205$).

Cut-off scores: The cut-off score indicating T-score elevations that are clinically significant are set at > 89T for the Adjustment scale, meaning that adjustment was within the normal range for all subgroups.

High risk group analysis: A total of 43 children (10%) had scores indicating a moderate or high risk of adjustment problems. There were no differences associated with gender ($\chi^2 = .04$, $p > .05$) or cleft visibility ($\chi^2 = .05$, $p > .05$). However, while only 3.2% ($n = 10$) of the children with a cleft only were in the high risk group, this was the case for 28% ($n = 33$) of the children with a cleft and an additional condition ($\chi^2 = 58.11$, $p < .001$).

Total difficulties score (SDQ)

2×2×2 ANOVA: As can be seen in Table 1, only one main effect was found on self- and parent reports, highlighting the risk of more psychological difficulties in children with an additional condition (EMM = 13.1, SE = .51) when compared to children with a cleft only (EMM = 9.5, SE = .42; F (1, 288) = 29.75, $p < .001$; $\eta^2 = 0.092$). The same effect was found in parent reports (Cleft only: EMM = 5.8, SE = .45; Cleft +: EMM = 11.5, SE = .53; F(1,294) = 67.90, $p < .001$). Effect size was large ($\eta^2 = 0.182$).

Reference group comparisons: On self-reports, girls with a cleft without an additional condition had similar scores (M = 9.8, SD = 4.9) to girls from the reference group (M = 10.1, SD = 5.1; t (1431) = 0.51, $p > .05$). The same was found in parent reports (Cleft: M = 6.1, SD = 4.8; Ref.gr.: M = 5.7, SD = 4.8; t (4121) = 0.56, $p > .05$). Boys with a cleft and no additional condition had less psychological adjustment problems on both self- (M = 9.2, SE = 4.9) and parent reports (M = 5.5, SD = 4.4) than boys from the reference group (Self-reports: M = 10.3, SD = 5.2; t (1560) = 2.10, $p < .05$; $d = -.22$; Parent reports: M = 6.6, SD = 5.2; t (4180) = 2.15, $p < .05$; $d = -.23$).

High risk group analysis: According to self-reports, 40 children (13.7%) were at high risk of adjustment difficulties, while 33 children (11.1%) were identified according to the parent reports. The only significant background factor was the presence of an additional condition. According to self-reports, 7.7% (n = 14) of the children with a cleft and no additional condition were in the high risk group, while parent reports identified 2.7% (n =5) children at high risk. In the group of children with an additional condition, approximately 25% were in the high risk group according to self-reports (n = 26; χ^2 = 15.45, p < .001) and parent reports (n = 28; χ^2 = 33.29, p < .001).

Cognitive Function

Cognitive function was measured by the Intellectual Screening scale from the PIC. In addition, two measures from the PIC and the SDQ provided information about problems with attention and/or hyperactivity, and were included as a measure of potential cognitive difficulties.

Intellectual Screening (PIC)

2×2×2 ANOVA: There were no interactions and two main effects (Table 1). As could be expected, children with an additional condition had higher scores on the Intellectual Screening scale, F(1, 436) = 268.27, p < .001), indicating more cognitive problems (EMM = 86.0, SE = 1.57) than children with a cleft only (EMM = 55.7, SE = .98). Effect size was very large (η^2 = 0.360). The second significant main effect was that children with a CP had more cognitive problems (EMM = 72.8, SE = 1.29) than children with a visible cleft (EMM = 68.9, SE = 1.32; F(1, 436) = 4.47, p < .05; η^2 = 0.006).

Cut-off scores: The cut-off score indicating elevations that are clinically significant are set at > 59T for the Intellectual Screening subscale. Hence, mean scores were above the clinical range for boys and girls, and irrespective of cleft visibility, when analyses were performed

without taking the presence of an additional condition into account. However, the children with a cleft and no additional condition had mean scores within the normal range irrespective of gender or visibility of cleft.

High risk group analysis: A total of 73 children (16.7%) were identified at high risk for cognitive problems according to the Intellectual Screening scale of the PIC. Within this group, 23.4% (n = 39) of children had a non-visible cleft compared to 12.6% (n = 34) of the children with a visible cleft ($\chi^2 = 8.48$, $p < .01$). Only 2.5% (n = 8) of the children with a cleft only were at high risk, in contrast to as many as half (53.3%; n = 65) of the children with a cleft and an additional condition ($\chi^2 = 162.22$, $p < .001$). Gender did not vary within the high risk group ($\chi^2 = .95$, $p > .05$).

Hyperactivity (PIC)

2×2×2 ANOVA: As can be seen in Table 1, there were two significant 2-way interactions, one between gender and an additional condition ($F(1,435) = 4.35$, $p < .05$), the other one between cleft visibility and an additional condition ($F(1,435) = 3.91$, $p < .05$). The patterns of these interactions were that the impact of an additional condition on problems with hyperactivity seemed to be stronger for the girls than for the boys, while the opposite pattern was the case in children without an additional condition. In addition, the impact of an additional condition was stronger in children with CLP than in children with CP. Effect sizes were small for both interactions ($\eta^2 < 0.010$), hence the details of the ANOVA are not reported in further detail.

There were two main effects. As could be expected, children with an additional condition had higher scores (EMM = 53.2, SE = 1.00) than children with a cleft only (EMM = 45.8, SE = .63; $F(1, 436) = 38.76$, $p < .001$; $\eta^2 = 0.360$) on the Hyperactivity scale. The second main effect indicated that children with CLP had more problems with hyperactivity (EMM = 51.0,

SE = .85) than children with CP (EMM = 48.0, SE = .83; F(1, 435) = 6.70, $p < .05$). This was probably associated with the interaction effect between cleft visibility and the presence of an additional condition. However, effect size was small ($\eta^2 = 0.006$).

Cut-off scores: The cut-off score indicating elevations that are clinically significant are set at > 59T for the Hyperactivity subscale, meaning that although statistics indicated significant differences between subgroups, mean scores were still within the normal range for all groups.

High risk group analysis: A total of 18 children (4.2%) were identified at high risk for problems with attention and hyperactivity. There were no gender differences in the high risk group ($\chi^2 = .93$, $p > .05$), and no differences related to cleft visibility ($\chi^2 = .00$, $p > .05$). Among the children with a cleft without an additional condition, only 1.3% ($n = 4$) had scores indicating high risk, while this was the case for 11.8% (n = 14) of the children with an additional condition ($\chi^2 = 23.75$, $p < .001$).

Attention and Hyperactivity (SDQ)

2×2×2 ANOVA: There were no interactions and one main effect on self-reports, while parent reports pointed to two main effects (Table 1). Children with a cleft and an additional condition expectedly had more problems with attention and/or hyperactivity (Self-reports: EMM = 5.0, SE = .21; Parent reports: EMM = 4.6, SE = .24) than children with a cleft only (Self-reports: EMM = 3.7, SE = .17; F(1, 288) = 21.27, $p < .001$; $\eta^2 = 0.075$; Parent reports: EMM = 2.3, SE = .20; F(1, 294) = 55.56, $p < .001$; $\eta^2 = 0.157$). The second main effect was found in parent reports only: boys had more problems with attention and/or hyperactivity (EMM = 3.8, SE = .21) than girls (EMM = 3.1, SE = .23; F(1, 288) = 4.77, $p < .05$). Effect size, however, was small ($\eta^2 = 0.013$).

Reference group comparisons: Girls with a cleft and no additional condition (M = 3.4, SD = 1.98) had similar scores as girls from the reference group on self-reports (M = 3.5, SD = 2.0; *t*

(1431) = 0.44, $p > .05$) and on parent reports (Cleft: M = 2.3, SD = 2.3; Ref.gr.: M = 2.2, SD = 2.0; t (4154) = 0.44, $p > .05$). The same was the case for the boys on self-reports (M = 4.0, SD = 2.1), as compared to those from the reference group (M = 3.8, SD = 2.1; t (1561) = 0.95, $p > .05$). The parents of boys with a cleft, on the other hand, reported significantly less problems with attention and hyperactivity (M = 2.5, SD = 2.07) than parents from the reference group (M = 3.0, SD = 2.4; t (4180) = 2.12, $p < .05$; d = -.22).

High risk group analysis: Cut-off scores identified 44 children (15.1%) at high risk for hyperactivity problems on self-reports, and 43 children (14.4%) according to parent reports. There were no gender differences (Self-reports: $\chi^2 = 1.42$, $p > .05$; Parent reports: $\chi^2 = 1.03$, $p > .05$), nor differences related to cleft visibility (Self-reports: $\chi^2 = 2.32$, $p > .05$; Parent reports: $\chi^2 = .22$, $p > .05$). As expected, there were significantly more children with a cleft and an additional condition in the high risk group (Self-reports: 24.1%, $n = 26$; Parent reports: 27.8%, $n = 32$) compared to children with cleft only (Self-reports: 9.8%, $n = 18$, $\chi^2 = 10.73$, $p < .01$; Parent reports: 6.0%, $n = 11$; $\chi^2 = 27.22$, $p < .001$).

Behavioural conduct

Behavioural conduct was measured through the Withdrawal scale (PIC) and the Conduct problems subscale (SDQ).

Withdrawal (PIC)

2×2×2 ANOVA: Analyses revealed no interactions and one main effect (Table 1). Children with an additional condition had higher scores on the Withdrawal scale (EMM = 54.2, SE = .89) than in cases of a cleft only (EMM = 51.1, SE = .55; F(1, 436) = 8.92, $p < .01$). Effect size was small ($\eta^2 = 0.020$).

Cut-off scores: The cut-off score indicating clinically significant elevations are set at > 69T for the Withdrawal subscale, meaning that mean scores were below the clinical range for all subgroups.

High risk group analysis: There were only two children (0.2%) at high risk for withdrawal difficulties according to the PIC. They were both boys, one with a non-visible cleft and no additional condition, the other one with a visible cleft and an associated condition.

Conduct problems (SDQ)

2×2×2 ANOVA: There was one main effect (Table 1). Children with a cleft and an additional condition had more conduct problems (Self-reports: EMM = 1.9, SE = .15; Parent reports: EMM = 1.7, SE = .14) than children with a cleft only (Self-reports: EMM = 1.5, SE = .12; $F(1, 288) = 5.30$, $p < .05$; $\eta^2 = 0.017$; Parent reports: EMM = 1.0, SE = .12; $F(1, 295) = 13.78$, $p < .001$; $\eta^2 = 0.044$).

Reference group comparisons: Girls with a cleft and no additional condition had similar scores as girls from the reference group on self-reports (Cleft: M = 1.5, SD = 1.35; Ref.gr.: M = 1.4, SD = 1.31; t (1431) = 0.66, $p > .05$) and parent reports (Cleft: M = 1.0, SD = 1.19; Ref.gr.: M = 1.1, SD = 1.4; t (4154) = 0.63, $p > .05$). The same was the case for boys on parent reports (Cleft: M = 1.1, SD = 1.29; Ref.gr.: M = 1.0, SD = 1.2; t (4180) = 0.84, $p > .05$). On self-reports, boys with a cleft reported significantly less conduct problems (M = 1.5, SD = 1.48) than the reference group (M = 2.0, SD = 1.74; t (1561) = 3.36, $p < .001$; $d = -.31$).

High risk group analysis: Cut-off scores identified 17 children (5.8%) at high risk for conduct problems according to self-reports and 26 children (8.7%) according to parent reports. Self-reports identified more boys (8.3%, $n = 14$) than girls (2.4%, $n = 3$; $\chi^2 = 4.49$, $p < .05$), while gender was non-significant in parent reports ($\chi^2 = .63$, $p > .05$). There were no differences related to cleft visibility ($\chi^2 = 2.69$ and .05, $p > .05$). Self-reports did not identify children

with an additional condition as at risk for conduct problems ($\chi^2 = .77$, $p > .05$), while parent reports did (5.5% vs. 13.9%; $\chi^2 = 6.33$, $p < .05$).

Social experiences

Social experiences were measured by the CEQ and the Peer problems subscale (SDQ).

Child Experience Questionnaire (CEQ)

2×2×2 ANOVA: There were no interactions and only one main effect (Table 1): children with a cleft and an additional condition reported less positive social experiences (EMM = 2.4, SE = .03) than children with a cleft only (EMM = 2.6, SE = .02; $F(1, 592) = 26.99$, $p < .001$; $\eta^2 = 0.043$).

Lack of norms and reference group: As far as the authors are aware, 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 (< 10th percentile). The high risk group consisted of 70 children (11.8%). The presence of an additional condition was the only significant risk factor (8.3%, $n = 34$ vs. 19.5%, $n = 36$; $\chi^2 = 15.22$, $p < .001$). There were no gender differences ($\chi^2 = 1.02$, $p > .05$), and no differences related to cleft visibility ($\chi^2 = .28$, $p > .05$).

Peer problems (SDQ)

2×2×2 ANOVA: There were no interactions and only one main effect on self-reports and parent reports (Table 1). Children with a cleft and an additional condition reported more peer problems (Self-reports: EMM = 2.6, SE = .17; Parent reports: EMM = 2.5, SE = .17) than children with a cleft only (Self-reports: EMM = 1.8, SE = .14; $F(1, 288) = 11.13$, $p < .01$;

Parent reports: EMM = 1.0, SE = .15; F(1, 295) = 46.11, $p < .001$). Effect sizes were small on self-reports ($\eta^2 = 0.039$), and large on parent-reports ($\eta^2 = 0.135$).

Reference group comparisons: Compared to reference groups, girls with a cleft and no additional condition reported the same level of peer problems (M = 1.9, SD = 1.7) as girls from the reference group on self-reports (M = 1.9, SD = 1.7; t (1431) = 0.00, $p > .05$) and parent reports (Both groups: M = 1.1, SD = 1.6; t (4154) = 0.00, $p > .05$). Boys with a cleft reported significantly less peer problems (M = 1.7, SD = 1.5) than the reference group on self-reports (M = 2.1, SD = 1.8; t (1561) = 2.23, $p < .05$; d = -.24) and on parent reports (Cleft: M = .8, SD = 1.3; Ref.gr.: M = 1.3, SD = 1.7; t (4180) = 2.99, $p < .001$; d = -.33).

High risk group analysis: Cut-off scores identified 34 children (11.7%) at high risk for peer problems according to self-reports and 47 children (15.8%) according to parent reports. There were no gender differences (Self-reports: $\chi^2 = .05$, $p > .05$; Parent reports: $\chi^2 = .05$, $p > .05$), and no difference related to cleft visibility ($\chi^2 = .01$, $p > .05$; $\chi^2 = 1.67$, $p > .05$). There were more children with an additional condition in the high risk group (Self-reports: 17.6%, n = 19; Parent reports: 31.3%, n = 36) than in cases of a cleft only (Self-reports: 8.2%, n = 15; $\chi^2 = 5.81$, $p < .05$; Parent reports: 6.0%, n = 11; $\chi^2 = 34.01$, $p < .001$).

Emotional Adjustment

Information about emotional adjustment was measured through the Depression and Anxiety scales of the PIC, and the Emotional difficulties scale of the SDQ, self- and parent reports.

Depressive Symptoms and Anxiety (PIC)

2×2×2 ANOVA: Analyses revealed no interactions and only one main effect (Table 1). Children with a cleft and an additional condition had more problems with depression (EMM = 58.0, SE = 1.10) than children with a cleft only (EMM = 50.4, SE = .69; F(1, 435) = 34.64, $p < .001$). The same was the case for anxiety symptoms (Cleft +: EMM = 59.2, SE = 1.07;

Cleft only: EMM = 52.3, SE = .66; F(1, 435) = 30.63, $p < .001$). Effect sizes were of medium range for depressive symptoms ($\eta^2 = 0.074$) and anxiety ($\eta^2 = 0.065$).

Cut-off scores: Cut-off scores that are clinically significant are set at > 69T for the Depression and Anxiety subscales, meaning that although statistics indicated significant differences between subgroups, mean scores were still within the normal range for all groups.

High risk group analysis: There were 15 children (3.5%) at high risk for depression and 10 (2.3%) at high risk for anxiety-related conditions. There were no differences related to cleft visibility ($\chi^2 = .49$ and .59, $p > .05$, respectively), and no gender differences ($\chi^2 = 2.30$ and .55, $p > .05$) in the high risk group. There were significantly more children with an additional condition (10.3%, $n = 12$ and 5.7%, $n = 7$) than children with a cleft only (1.0%, $n = 3$; $\chi^2 = 21.85$, $p < .001$ and 1.0%, $n = 3$; $\chi^2 = 8.97$, $p < .01$).

Emotional difficulties (SDQ)

2×2×2 ANOVA: There was one interaction in self-and parent reports, two main effects in self-reports, and one main effect in parent reports (Table 1). On self-reports, the pattern of the interaction was that while the girls with a cleft had rather high scores whether they had an additional condition or not, the impact of an additional condition seemed more important in boys ($F(1,288) = 3.95$, $p < .05$). In parent reports, the interaction was related to gender and cleft visibility ($F(1,288) = 8.80$, $p < .01$). Girls with a visible cleft reported less emotional difficulties than girls with a non-visible cleft, while the opposite was the case for boys. However, effect sizes were small for both interactions ($\eta^2 < 0.017$).

The main effects in self-reports involved gender and the presence of an additional condition. Girls reported more emotional difficulties (EMM = 3.4, SE = .22) than boys (EMM = 2.8, SE = .19; $F(1,288) = 4.35$, $p < .05$). Effect size, however, was small ($\eta^2 = 0.013$). The other main effect was once again related to the presence of an additional condition (Cleft: EMM =

2.5, SE = .19; Cleft+: EMM = 3.7, SE = .23; F (1,288) = 16.35, $p < .001$). There was only one main effect in parent reports, associated with the presence of an additional condition (F (1,295) = 23.96, $p < .001$). Effect sizes were within the medium range on self-reports (η^2 = 0.046) and parent reports ($\eta^2 = 0.069$).

Reference group comparisons: Girls with a cleft and no additional condition (M = 3.0, SD = 2.2) reported similar levels of emotional problems as girls from the reference group on self-reports (M = 3.0, SD = 2.2; t (1431) = 0.00, $p > .05$), and had more emotional problems according to parent reports (Cleft: M = 1.8, SD = 1.9; Ref.gr.: M = 1.4, SD = 1.8; t (4154) = 1.95, $p = .051$; $d = .22$). However, this difference was not statistically significant. Boys with a cleft (M = 2.2, SD = 2.1) reported similar levels of emotional difficulties as the reference group on self-reports (M = 2.2, SD = 2.1; t (1561) = 0.00, $p > .05$) and parent reports (Cleft: M = 1.2, SD = 1.4; Ref.gr.: M = 1.2, SD = 1.7; t (4180) = 0.00, $p > .05$).

High risk group analysis: There were 43 children (14.8%) at high risk for emotional problems according to self-reports, and 38 children (12.8%) according to parent reports. Self-reports revealed more girls (20.3%, $n = 25$) than boys (10.7%, $n = 18$) in the high risk group (χ^2 = 5.21, $p < .05$), while this difference was not significant in the parent reports ($\chi^2 = .53$, $p > .05$). Self-reports also identified more children with a CP (21.0%, $n = 22$) in the high risk group than children with CLP (11.4%, $n = 21$; $\chi^2 = 4.89$, $p < .05$), while parent reports did not (χ^2 = 2.28, $p > .05$). While 10.4% ($n = 19$) of the children with a cleft only were found in the high risk group, this was the case for 22.2% ($n = 24$) of the children with an additional condition ($\chi^2 = 7.56$, $p < .01$). Approximately the same pattern was found in parent reports (5.5%, n = 10 vs. 24.3%, $n = 28$; $\chi^2 = 22.63$, $p < .001$).

Satisfaction with appearance

Satisfaction with appearance was measured using the SWA designed by the Psychology Special Interest Group of the CFSGBI.

2×2×2 ANOVA: As can be seen in Table 1, analyses revealed only one main effect, children with an additional condition reporting less satisfaction with appearance (EMM = 8.1, SE = .11) than children with a cleft only (EMM = 8.5, SE = .07; F (1,676) = 9.23, $p < .01$). However, effect size was small ($\eta^2 = 0.014$).

In order to further explore whether cleft visibility could affect satisfaction with specific parts of the face, a new variable was computed that included the items from the SWA known to be potentially affected by a cleft: the face, nose, lip, teeth, speech, and the child's subjective evaluation of cleft visibility. Mean scores were computed and the same analyses as described above were performed. No significant 2-way interactions were found, but there were two main effects (Table 1). Not surprisingly, children with a visible cleft reported less satisfaction on cleft affected areas of the face (EMM = 7.5, SE = .11) than children with a non-visible cleft (EMM = 8.2. SE = .12; F (1,676) = 17.90, $p < .001$). The second significant difference was related to the presence of an additional condition (Cleft+: EMM = 7.5, SE = .11; Cleft: EMM = 8.2, SE = .12; F (1,676) = 6.49, $p < .05$). However, effect sizes were small for both main effects ($\eta^2 < 0.026$).

Lack of norms and reference group: As far as we know, no published 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: Percentile analyses revealed that a mean of 6.18 or lower was indicative of high risk for dissatisfaction with total appearance (< 10th percentile). A total of 66 children (9.7%) were found within the high risk group. There were no gender differences ($\chi^2 = .74$, $p > .05$), no differences related to visibility of cleft ($\chi^2 = .44$, $p > .05$), and no

differences regarding the presence or absence of an additional condition ($\chi^2 = 2.39$, $p > .05$) between the high risk and the non-risk group.

Risk groups across measures

In order to compare risk groups across measures, five new variables were created[2]. These five variables recorded the children that 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 that had been used initially. In addition, children reporting scores within the borderline range were identified and recorded. Hence, as an example, children at risk for depressive symptoms and anxiety (PIC), and/or those identified at risk for emotional difficulties (SDQ) were recorded in the new variable named "Emotional adjustment". An overview of the frequency of children 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 2.

In total, 20.5% ($n = 146$) were found to be at high risk for cognitive and/or attention difficulties, 5.6% ($n = 40$) at high risk for behavioural problems, 17.7% ($n = 114$) at high risk for social difficulties, 12.1% ($n = 86$) at high emotional risk, and 9.8% ($n = 66$) were at high risk for dissatisfaction with appearance. As can be seen in Table 2, frequencies of children within the borderline range varied between 7.5 and 26% of the total sample, depending on the domain of risk.

A total of 32.9% of the children ($n = 175$) belonged to none of the risk groups, while 21.4% ($n = 114$) had scores on the borderline range in one domain only. When categorising the children into normal/borderline versus high risk groups, 62.4% of the children ($n = 333$) belonged to none of the high risk groups, while 22.9% ($n = 122$) were at high risk in one

[2] General adjustment (PIC) and the Total difficulties score of the (SDQ) are both based on the instruments' subscales, and were therefore not included in further analyses of high risk groups.

group, 10.5% (n = 56) were at risk in two groups, 3.0% (n = 16) in three groups, while six children (1.1%) were found to be at high risk on all five domains of risk.

Correlations between the five risk groups were calculated. Most correlations were significant, and varied from no associations to moderate associations. The strongest association was found between social and emotional risk (r = .38, n = 598, p < .001). The other correlations were, in order of strength of association: emotional and behavioural risk (r = .35, n = 708, p < .001), emotional and cognitive risk (r = .31, n = 711, p < .001), social and behavioural risk (r = .28, n = 596, p < .001), behavioural and cognitive risk (r = .28, n = 708, p < .001) and cognitive and social risk (r = .23, n = 598, p < .001). The remaining four correlations were weak or non-significant: appearance and social risk (r = .18, n = 572, p < .001), appearance and cognitive risk (r = .12, n = 631, p < .01), appearance and emotional risk (r = .07, n = 620, p > .05) and appearance and behavioural risk (r = 0.03, n = 633, p > .05).

Psychometric properties

Correlations across measures and informants

Calculations of convergent validity and levels of agreement between child and parent reports are presented in Table 3. Correlations between the CEQ and the Peer problems subscale of the SDQ were moderate, as was the case for levels of agreement between child and parent reports for the SDQ. Correlations were similar or higher than previously reported (Goodman, 2001; Van Roy et al, 2010).

Convergent validity was also calculated between the PIC and the SDQ. However, since the SDQ had replaced the PIC during the period of data collection, information from both measures existed only for 25-30 participants. Correlations showed associations ranging from r = -.10 to .80, the lowest being across informants (child vs. parent on same adjustment domain), the highest within informants (child vs. child and parent vs. parent). However, the

sample was estimated to be too small for a test of convergent validity, and results are hence not reported in more detail.

Internal consistency

The PIC and the SDQ are both validated measures, while the CEQ and the SWA are not. Internal reliability was calculated for all measures and is reported in Table 4. Psychometric properties varied significantly across and within measures, irrespective of whether they have been validated in the past or not. Reliability was acceptable for the CEQ, suggesting its usefulness as a total measure of social experiences. While some subscales of the SDQ and the PIC had good to excellent internal reliability, other subscales had poor or unacceptable internal reliability.

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 children with a cleft.

The prevalence of cognitive, behavioural, emotional, social, and appearance-related risk was significantly associated with the presence of an additional condition in all measures, while the effect of cleft visibility and gender seemed to be less important at age 10. Approximately 60% of the children were not at high risk in any of the adjustment domains. Less than 25% were at high risk in one domain only, while approximately 15% were at high risk in two or more domains of adjustment.

The strongest associations were found between social and emotional risk and social and behavioural risk. Although these associations were significant, the effects can only be

interpreted as small to moderate. Dissatisfaction with appearance did not seem to be associated with other psychological difficulties at this age. The results of the present study thus point towards risk and resilience as being domain-specific, rather than general.

Psychological functioning: The role of an additional condition

The risk of cognitive impairment, behavioural difficulties, emotional distress, psychosocial problems, and dissatisfaction with appearance in children born with a cleft was associated with the presence of an additional condition, while being non-related to visibility of cleft. The only exception was cognitive difficulties, which were more often associated with cleft palate, as demonstrated in the previous literature (Christensen and Mortensen, 2002; Swanenburg et al. 2003). However, effect size related to cleft type was weak, in contrast to a very large effect associated with the presence of an additional condition.

The results of the present study clearly confirm 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 could be at psychological risk. Approximately one third of the children had one or more conditions in addition to the cleft, and the presence of an additional condition was a strongly significant indicator of risk within all domains of adjustment. However, when comparing the results with comparison samples, mean scores were still within the normal or borderline range, in spite of being elevated compared to the children with a cleft only. When investigating high risk groups, the prevalence of children with an additional condition ranged from 10 to 50% as compared to 1 to 10% of children with a cleft only. These results are not surprising, since several conditions included in the present sample are well-known to be associated with risk for psychological and/or cognitive problems, such as 22q11.2 (Green et al., 2009), language and reading difficulties (Goodyer, 2000), or AD/HD (Spencer, 2006; Wehmeier et al., 2010).

When excluding the children with an additional condition, less than 3% of the children in the total sample had cognitive problems that were clinically significant, while 5-10% had problems related to attention and/or hyperactivity. This is in contrast to findings reporting that approximately 46% of children with cleft have a learning disability (Broder et al., 1998), while it is similar to the frequency that was found in the group of children with a cleft and an additional condition in the present study. The current findings therefore highlights the importance of evaluating whether the cognitive problems that are often reported in cleft samples could primarily or partly be associated with the presence of undiagnosed or unidentified additional conditions, rather than being a direct consequence of the cleft itself.

Conversely, a growing literature investigates neurological aspects of cleft lip and palate (Nopoulos et al., 2007; Richman et al., 2012), identifying structural brain differences which could explain the presence of cognitive difficulties in children with non-syndromic clefts. One of the challenges for future research would be to disentangle the complex relationship between cleft-specific problems and those related to the presence of other co-morbid conditions. The comorbidity of clefts and other conditions in some individuals could suggest a genetic double association as an indication of syndromes not yet identified (Richman and Ryan, 2003). The results of the present study further demonstrate the importance of identifying not only children with syndromes and severe developmental difficulties, but also those with less impacting conditions, since psychological problems within different domains of adjustment have been found across groups (Feragen and Stock, 2014). The wide range of different associated conditions should bring about the question of which co-morbid diagnoses are excluded, and consequently which associated problems are likely to remain in cleft samples (Feragen et al., in press). Further research is also needed in order to explore potential differences between subgroups of additional conditions in terms of psychological risk, and

whether the number of additional conditions adds risk for psychosocial adjustment difficulties.

Risk and Protection

Boys with a cleft only showed more positive adjustment on several domains compared to same-aged boys from the reference groups, while girls with a cleft had similar scores as girls from the general population. In addition, almost 60% of the children within the sample had scores within the normal/borderline range on all domains of adjustment. This could indicate the presence of protective factors that counteract the consequences of potential risk. The results suggest that most children with a cleft cope well, in spite of specific challenges that are known to be associated with living with a visible difference. Further, the lack of strong associations between the risk groups suggest that risk seem to be domain-specific, and not general in children with a cleft. This could indicate that interventions tailored within specific domains of risk may be efficient for most children with this condition. Of the five domains, only social and emotional risk and emotional and behavioural risk were found to be associated at a level that was considered clinically significant. However, the magnitude of these associations was moderate. Additionally, being dissatisfied with subjective appearance at age 10 was not associated with emotional, behavioural or psychosocial difficulties. Interestingly, a similar finding was reported in adults with a cleft (Roberts and Mathias, 2012), while other studies have pointed to the importance of subjective appearance evaluations for psychological adjustment in older participants (Feragen et al., 2010; Mani et al., 2013). For the present age group, the findings could hence point towards the effectiveness of interventions which taps into specific domains of risk, such as social skills training, cognitive-behavioural interventions, or interventions directed towards reducing emotional distress (Robinson et al., 1996; Maddern and Owen, 2004; Kapp-Simon et al., 2005; Bessell et al., 2012), when problems have been identified within these specific areas of adjustment. Alternatively,

731 interventions could aim at strengthening resilience in other domains, in order to reduce risk.
732 For the children at risk in several domains however (approximately 15% of the sample in the
733 current study), interventions should be delivered at a broader level, in order to capture the
734 potential associations between several domains of adjustment.

735 Due to the study's retrospective and cross-sectional nature, the causal links between
736 associations could not be determined. Behavioural difficulties were associated with all other
737 domains of adjustment to a moderate degree. Since less than 6% of the total sample had
738 behavioural difficulties, conduct problems seem to be a consequence of social, emotional
739 and/or cognitive risk in a subgroup of children, rather than behavioural difficulties being
740 generally associated with having a cleft. Further, the association between emotional and
741 social adjustment could suggest that emotional difficulties are a consequence of negative
742 psychosocial experiences, as have been shown in the general population (Roberts and
743 Mathias, 2012; Guederey et al., 2014), and in cleft research (Murray et al., 2010). However,
744 previous literature has also shown that emotional difficulties may affect the child's ability to
745 form social relationships (Graber, 2004). In the present study, the domain related to social
746 experiences was the one revealing the highest frequency of risk in children with a cleft,
747 without a corresponding prevalence of emotional risk. If social risk predisposes to emotional
748 problems, more children could have been expected to be at emotional risk in the present
749 study. Hence, in spite of a relatively high number of children at social risk, significantly fewer
750 children were at high risk within the other domains of adjustment, which could indicate the
751 presence of potential protective factors in the sample, such as positive self-concepts and
752 cognitive processes (Moss, 2005; Rumsey and Stock, 2013), close friendships and positive
753 social experiences (Feragen et al., 2010), efficient coping strategies and social skills (Kapp-
754 Simon et al., 2005; Baker et al., 2009; Berger and Dalton, 2011), and positive emotional
755 adjustment (Feragen et al., 2009). Ultimately, Masten's conceptualization of resilience (2001)

suggests that it may not necessary to search for extraordinary mechanisms in this population because the ‘‘ordinary magic’’ is the child’s capacity for positive and normal adjustment in spite of challenging experiences.

Social disadvantage due to the visible difference have been reported previously (Murray et al., 2010), and has been supported by neuropsychological findings related to social function (Canady et al., 2007). However, the current findings did not indicate that the children in this study were at social and emotional risk because of cleft visibility. Such findings address the need for research to identify other risk factors in this population, and to acknowledge positive adjustment factors (Egan et al., 2011; Roberts and Mathias, 2012), in order to capture the complexity of adjustment to a visible difference (Stock et al., 2013). Longitudinal studies are ultimately needed in order to address the directionality of associations, and whether risk groups would be found within the same adjustment domains in later developmental stages.

Gender differences

Differences between boys and girls at age 10 were investigated within the cleft sample, and in comparison to the reference groups. Within the cleft sample, gender differences were found in relation to emotional difficulties and problems with attention. When comparing the cleft sample to the reference group, gender differences indicated more positive general adjustment in boys with a cleft, in addition to fewer problems related to attention and peers on the SDQ.

Within the cleft sample, boys were more at risk for problems with attention than girls according to parent reports, while girls were at greater emotional risk on self-reports. Such gender differences are in line with findings from the general population (Rønning et al., 2004; Van Roy et al., 2006; Van Roy et al., 2010). However, when comparing the cleft sample with the reference group, parent-reports indicated that boys with a cleft had less attention problems and less social problems than the reference group. Parents of girls from the cleft sample

780 reported more emotional difficulties than girls from the reference groups. However, this
781 difference was not statistically significant, and the effect size was small.

782 Interestingly, interactions between gender, cleft visibility and the presence of an additional
783 condition were found for problems of attention and hyperactivity (PIC) and for emotional
784 difficulties (SDQ). These findings indicated that the presence of an additional condition had a
785 greater impact on problems of attention and hyperactivity in girls than in boys, and more on
786 children with CLP. Regarding emotional distress, the impact of an additional condition
787 seemed greater for boys than girls. A second interaction pointed to more emotional problems
788 in boys with CLP and girls with CP, than in girls with CLP and boys with CP. These findings
789 could indicate that gender-related risk varies depending on whether the child has an additional
790 condition or not, and possibly additionally related to cleft type, once again highlighting the
791 importance of careful identification of subgroups of children with a cleft.

792 The reported findings from the present study need to be viewed in light of the questionable
793 internal reliability that was reported for a number of SDQ subscales, including self-reports of
794 attention, peer problems and emotional difficulties, as well as parent-reports of peer problems
795 and emotional difficulties. However, the Total difficulties score on the SDQ demonstrated
796 good reliability, and thus the overall conclusion can be drawn that boys report less adjustment
797 problems than the reference groups. This finding is also in line with a previous study that
798 pointed to processes of resilience in adolescent boys with a visible difference (Feragen et al.,
799 2010). Further studies are needed in order to investigate whether there could be gender-
800 specific protective factors at work.

801 The present study included only children aged 10, in contrast to many cleft samples often
802 including children from a wide age range, complicating the interpretation of findings and
803 comparisons between studies. Since social challenges and psychological difficulties have

been shown to increase from childhood to adolescence, especially in girls (Dekker et al., 2007; Smolak, 2012; Snyder and Pope, 2010), results from samples with wide age ranges may be imprecise and gender differences be blurred by differences related to age. Clearly defined age groups are needed to explore adjustment across different developmental stages. Gender differences in the general population point to the importance of gender-specific results also in the cleft literature. In order to be able to explore this, large samples are needed; a factor that probably explains the choices related to age and gender made in many studies.

Generic vs. specific measures in cleft clinics

As previously discussed, there is an on-going dialogue about whether to use generic or specific measures in cleft research and clinics. The present study was primarily based on data collected using generic measures, while the outcome variable measuring satisfaction with appearance was cleft-specific. Interestingly, this was also the only measure that indicated more negative findings for children with a visible cleft compared to those with a non-visible cleft, when including only the measure's cleft-specific items.

The fact that cleft visibility did not affect the outcome measures could be explained in different ways. One interpretation is that cleft visibility in itself is not the main issue for psychological adjustment, as has been demonstrated by several recent studies (Moss, 2005; Appearance Research Collaboration, 2009; Feragen et al., 2010). Another interpretation could be that generic measures are not sensitive or specific enough to actually highlight existing difficulties or condition-specific challenges. A third interpretation could be that children with a cleft, in spite of, or because of the challenges involved in their condition, still develop an ability to cope with their condition, resulting in positive adjustment. The lack of strong associations between the different risk groups, and the positive adjustment findings in comparison to reference groups, could support this final hypothesis. Further, the lack of associations between dissatisfaction with appearance and other domains of risk, suggests that

829 children aged 10 with a visible difference who are dissatisfied with their appearance are not
830 necessarily at risk for emotional or social distress, in contrast to what has been demonstrated
831 in the general population (see Rumsey, 2008 for a review). The lack of associations between
832 dissatisfaction with appearance and other domains of risk may be specific to this age group,
833 and stronger associations between domains of risk could be expected in adolescents and
834 young adults (Dekker et al., 2007; Smolak, 2012).

835 There is a need for both generic and specific measures if we are to fully understand the
836 complexities of adjustment in children and young people with a cleft. Clinical psychologists
837 in cleft teams need to have reliable and valid measures that help them to identify children at
838 risk, both in general terms and in relation to those struggling with cleft-specific challenges.
839 The present study highlights that while children with a cleft in general have good
840 psychological health, some subgroups are more at risk when it comes to cognitive and
841 behavioural functioning, social experiences, emotional adjustment and appearance-related
842 satisfaction.

843 *Psychometrics: Convergent validity, agreement between informants and internal reliability*
844 All measures used in this study confirmed the presence of an additional condition as a risk
845 factor, while gender and cleft visibility did not seem to affect adjustment. These similarities
846 in findings were present irrespective of the measure's psychometric properties. Nevertheless,
847 the usefulness of any measure depends on its psychometric properties, such as validity and
848 reliability. As mentioned throughout this paper, some of the subscales, on the SDQ as well as
849 the PIC, were found to have questionable or even unacceptable psychometric properties. Low
850 internal consistency could indicate that results, such as those related to cognitive problems
851 and difficulties with attention, should be interpreted with caution. On the other hand, a recent
852 paper (McCrae et al., 2011) suggests that while Cronbach's alpha is useful as an indicator of
853 the degree to which constituent parts of a whole cohere, it appears to be of limited utility for

854 evaluating the validity of a scale. Unfortunately, the present study was not in the position to
855 assess the measures' validity, since participants had not completed instruments measuring
856 similar constructs. Hence, an interpretation of the results has to rely on other studies having
857 assessed the validity of the same subscales. Convergent validity has been shown in a number
858 of studies for the SDQ (Goodman, 2001; Van Roy et al., 2008) and the PIC (Wirt et al.,
859 1984). In the present study, measures of reliability were similar or better to those reported in
860 other studies for the SDQ (Goodman, 2001; van Roy et al., 2008; Stone et al., 2010) and the
861 PIC (Wirt et al., 1984). It has been argued that low internal reliability on the hyperactivity,
862 conduct, and peer problems subscales of the SDQ may be due to the positively worded
863 reverse-scored items, or may possibly also be related to the limited number of response
864 categories (Van Roy et al., 2008). In summary, questionable internal reliability on the SDQ
865 and the PIC may be counterbalanced by the many studies having evaluated the scales'
866 external and convergent validity (Wirt et al., 1984; Goodman, 2001; Van Roy et al., 2008).

867 Level of agreement between children and parents on the same subscales were calculated and
868 showed moderate associations, the lowest being emotional distress. Differences in self- and
869 parent reports have been described previously when using the SDQ, and the level of
870 agreement was similar or higher in the present cleft population (Goodman, 2001; Van Roy et
871 al., 2010). Higher agreement on measures of peer problems could be due to the parents'
872 capacity to observe and identify social problems due to their visibility in daily life, as
873 compared to emotional difficulties, which may not be apparent to anyone other than the
874 affected person. Differences between self- and parent-reports highlight the importance of
875 using as many informants as possible in order to shed light on the complexity of perceptions
876 of psychological adjustment.

877 In light of the findings of this study, a number of observations can be made with regard to the
878 clinical and research utility of the measures used. Although the PIC has been previously

validated (Troland, 1988; Wirt et al., 1984), and its psychometric properties appear to be good on a number of clinical scales, it has not been well used in other CL/P studies, complicating comparisons. Additionally, as far as known, the PIC has not been translated into a range of languages, which also limits its use for many cleft teams. Nevertheless, the PIC provides clinically useful findings since it is possible to categorise children according to risk groups on several psychological domains of adjustment, and would thus be useful for other studies to consider using it in the future. Since data were collected within the present study, this measure has been adapted into the more recent PIC-2 (Lachar and Gruber, 2002). The PIC-2's age range has been expanded to range from 5 to 19 years, providing the possibilities of longitudinal data within cleft cohorts.

The SDQ is user-friendly 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. Unfortunately, internal reliability in this and in other studies (Goodman, 2001; Rønning et al., 2004; Stone et al., 2010) has been shown to be poor, unacceptable or questionable for some subscales, such as the ones measuring conduct difficulties, peer problems and emotional difficulties. The subscale measuring problems with attention and/or hyperactivity had good reliability on parent reports, while self-reports at age 10 were questionable. Nonetheless, the Total difficulties score showed good reliability and correlated highly with the general adjustment scale from the PIC. 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 CEQ has been used in cleft research previously, but published norms are not available. Unfortunately, the measure has been used differently across studies and results are sometimes calculated in alternate ways, making meaningful comparisons more challenging. Although

the psychometric properties of the CEQ were considered acceptable within this study, the scale is more difficult to interpret without norms. Despite this, some cleft teams do find this measure clinically useful. Its associations with the peer problems subscale of the SDQ indicated good convergent validity.

The SWA has also been used in a number of cleft studies and was the only measure 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, and convergent validity could not be computed since other appearance-related measures were not used in the present data-set. Normative data have been reported to exist for a UK sample, and are reported in Berger and Dalton (2009). However, the age range includes children and adolescents, complicating comparisons, and most probably obscuring age-specific differences in satisfaction with appearance.

Strengths and Limitations

The main strength of the current study was its large and representative sample of eleven consecutive birth cohorts, presenting adjustment from a cognitive, behavioural, emotional, social and appearance-related perspective. This comprehensive approach allowed an investigation of whether different domains of risk and resilience could be working within the same individual, or whether risk was more general in nature, within a restricted age range, hence reducing the confound of age and/or developmental stages. Furthermore, the sample included children with an associated condition, raising awareness about potentially vulnerable subgroups. Results were based on data from both child and parent reports. Another strength was that both mothers and fathers contributed information, which is still rare in paediatric psychological research (Stock and Rumsey, in press). Additionally, results from the SDQ could be compared to same-aged reference groups from large national samples. Further, by

running a 2×2×2 ANOVA instead of several *t*-tests, the chances of Type I error were kept at 5%, and estimated marginal means were adjusted for the other variables in the model. Hence, more correct estimations of the variable's effect on outcome were provided.

Limitations of this study included the lack of control group for some of the outcome measures, and poor psychometric properties on a number of subscales. However, by discussing these issues in relation to the results, the limitations were partially counteracted. If future studies were able to provide this information it could help researchers and clinicians to understand more about the nature of the discrepancies that are often found across studies.

Due to its retrospective nature, the study was restricted by the measures that had been used during routine assessments. Hence, even if most areas of research were addressed that had been identified in recent systematic reviews and book chapters, some measures may not have been optimal in capturing specific issues of adjustment. As an example, cognitive risk may have been better assessed with tests of cognitive performance and abilities. Another limitation could be the lack of data for the children with severe developmental problems who did not go through the routine assessment, since they were not able to complete any of the measures used. Their presence in the sample would probably have impacted on the mean scores for most variables, increasing the findings related to risk in the group of children with an additional condition, and needs to be acknowledged. Further, adjustment to a visible difference involves a combination of psychological and societal factors that were not accounted for in the present study, such as individual characteristics, cognitive processes (such as attribution style or coping strategies), family factors and social support, in addition to socio-cultural factors. An additional variable of potential importance in children with a cleft is related to problems with speech, a variable which has been shown to be associated with social difficulties (Watterson et al., 2013). Unfortunately, speech outcomes other than the child's subjective satisfaction were not available in the present data set. Future research

954 should aim to include such information. Ultimately, longitudinal studies are needed in order
955 to understand how patterns of risk may vary from childhood to adolescence.

956 *Summary and conclusions*

957 The objectives of the present paper were to investigate whether there were associations
958 between different domains of risk at age 10 and to explore the usefulness of measures of
959 psychological adjustment across a range of domains. Approximately a third of the children
960 were not at risk on any adjustment measure, while another 20% were within the borderline
961 range on one domain only. The number of children at high risk in more than one domain of
962 adjustment was less than 15%, and few associations were found between risk groups.
963 However, emotional and social risk were more closely related than other risk groups.
964 Objective cleft visibility did not seem to be an important factor at age 10, and boys with cleft
965 appear to experience less overall adjustment difficulties than the reference groups. The
966 results seem to point to risk factors as well as potential protective factors in children with a
967 cleft lip and/or palate at age 10. Children with a condition in addition to a cleft were found to
968 be at higher risk across all measures. Findings from the present study therefore also point to
969 the importance of early screening and assessment of children born with a cleft, in order to
970 identify possible associated conditions and offer adapted and appropriate treatment and care.
971 Finally, this study has examined a number of measures pertaining to psychological adjustment
972 at age 10 in relation to clinical relevance and psychometric value.
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Table 1. Results from the 2×2×2 ANOVA's assessing the significance of gender, cleft visibility, and the presence of an additional condition at age 10 on all outcome variables.

Psychological adjustment		**Main effects and Interactions**	**F**	**R^2**
General adjustment	PIC (mother)	Gender Cleft visibility Additional condition	3.30 4.12* 116.95***	.25
Total score	SDQ (self-reports)	Gender Cleft visibility Additional condition	0.00 0.01 29.75***	.13
	SDQ (parent reports)	Gender Cleft visibility Additional condition	0.35 0.50 67.90***	.25
Cognitive function				
Intellectual Screening	PIC (mother)	Gender Cleft visibility Additional condition	1.98 4.47* 268.27***	.42
Hyperactivity	PIC (mother)	Gender Cleft visibility Additional condition Gender*Additional condition Cleft visibility*Additional condition	0.05 6.70* 38.76*** 4.35* 3.91*	.12
Attention/Hyperactivity	SDQ (self-reports)	Gender Cleft visibility Additional condition	1.76 0.16 21.27***	.10
	SDQ (parent reports)	Gender Cleft visibility Additional condition	4.77* 0.85 55.56***	.21
Behavioural conduct				
Withdrawal	PIC (mother)	Gender Cleft visibility Additional condition	.57 .07 8.92**	.03
Conduct problems	SDQ (self-reports)	Gender Cleft visibility Additional condition	0.83 0.94 5.30*	.06
	SDQ (parent reports)	Gender Cleft visibility Additional condition	1.20 1.77 13.78***	.07
Social experiences				
Social experiences	CEQ (self-reports)	Gender Cleft visibility Additional condition	0.72 0.06 26.99***	.06
Peer problems	SDQ (self-reports)	Gender Cleft visibility Additional condition	0.39 0.01 11.13**	.08
	SDQ (parent reports)	Gender Cleft visibility Additional condition	0.11 0.87 46.11***	.19
Emotional adjustment				
Depression	PIC (mother)	Gender Cleft visibility Additional condition	0.30 0.18 34.64***	.09
Anxiety	PIC (mother)	Gender Cleft visibility Additional condition	1.49 1.08 30.63***	.09
Emotional difficulties	SDQ (self-reports)	Gender Cleft visibility Additional condition Gender* Additional condition	4.35* 0.04 16.35*** 3.95*	.09
	SDQ (parent reports)	Gender Cleft visibility Additional condition	2.30 0.99 23.96***	.15

		Gender*Cleft visibility	8.80**	
Appearance satisfaction				
Satisfaction with appearance	SWA (self-reports)	Gender Cleft visibility Additional condition	1.82 1.16 9.23**	.02
Satisfaction with appearance Cleft-related items	SWA-cleft (self-reports)	Gender Cleft visibility Additional condition	3.23 17.90*** 6.49*	.04

Note: * $p < .05$; ** $p < .01$; *** $p < .001$. In order to simplify the Table, two- and three-ways interactions are only reported when significant.

Table 2. Risk groups across domains of psychological adjustment

		Normal range	**Borderline**	**High risk**
	n	**% (*n*)**	**% (*n*)**	**% (*n*)**
Cognitive risk	712	66.2 (471)	13.3 (95)	20.5 (146)
Behavioural risk	709	87.0 (617)	7.5 (53)	5.5 (40)
Social risk	644	56.7 (365)	25.8 (166)	17.5 (113)
Emotional risk	712	77.4 (551)	10.5 (75)	12.1 (86)
Appearance-related risk	675	75.6 (510)	14.7 (99)	9.8 (66)

Table 3. Associations between subscales within and across measures for the cleft sample: self-reports (S) and parent reports (P).

Measures compared		Informants			*n*	Pearson's *r*
General adjustment		S-S	S-P	P-P		
Total difficulties score (SDQ)	Total difficulties score (SDQ)		X		281	.45***
Cognitive function						
Intellectual Screening (PIC)	Hyperactivity (PIC)			X	436	.29***
Attention problems (SDQ)	Attention problems (SDQ)		X		281	.42***
Behavioural difficulties						
Conduct problems (SDQ)	Conduct problems (SDQ)		X		282	.32***
Social experiences						
Social experiences (CEQ)	Peer problems (SDQ)	X			247	.55***
Social experiences (CEQ)	Peer problems (SDQ)		X		247	.46***
Peer problems (SDQ)	Peer problems (SDQ)		X		282	.46***
Emotional adjustment						
Depressive symptoms (PIC)	Anxiety (PIC)			X	436	.84***
Emotional problems (SDQ)	Emotional problems (SDQ)		X		282	.28***
Satisfaction with appearance						
Satisfaction with appearance (SWA)	Cleft-related items (SWA)	X			621	.89***

Note:. ** $p < .01$, *** $p < .001$.

Table 4. Internal consistency (Cronbach's alpha) in the present study for the different measures.

Measure		**Subscales**	***n***	**α**	
Personality Inventory for Children	PIC	Adjustment	437	.81	Good
		Intellectual Screening		.61	Questionable
		Withdrawal		.57	Poor
		Depression		.83	Good
		Anxiety		.75	Acceptable
		Hyperactivity		.48	Unacceptable
Child Experience Questionnaire	CEQ		550	.73	Acceptable
Strengths and Difficulties Questionnaire	SDQ		280		
Self-reports		Total difficulties score		.78	Acceptable/Good
		Emotional difficulties		.68	Questionable
		Conduct difficulties		.47	Unacceptable
		Attention/Hyperactivity		.60	Questionable
		Social/Peer		.50	Poor
Parent-reports		Total difficulties score	289	.85	Good
		Emotional difficulties		.66	Questionable
		Conduct difficulties		.57	Poor
		Attention/Hyperactivity		.80	Good
		Social/Peer		.66	Questionable
Satisfaction with Appearance scales	SWA		632	.89	Good/Excellent