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

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36

**Abstract**

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

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

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

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

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

58

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

61

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

74 Although studies often aim to investigate the same areas of psychological adjustment, there is  
75 a clear discrepancy in the measures which are used (Klassen et al., 2012; Rumsey and Stock,  
76 2013), complicating comparisons between studies. In addition to the need for comparisons,  
77 there is a need to agree upon measures which would help researchers to discriminate clearly  
78 between those children with CL/P who cope well and those who may be at risk. One  
79 additional consideration in regard to choosing instruments is whether to use generic measures  
80 of psychological wellbeing or more condition-specific measures. While generic measures  
81 provide universal information that can be compared to reference groups and control groups,  
82 specific measures may be more sensitive to the aspects and challenges associated with a  
83 particular condition (Roberts and Shute, 2011). Although there is a probability that a  
84 combination of both types of measures would be most helpful, clear guidance is not available  
85 due to the current lack of consistency within research findings. Agreeing on measures is a  
86 cumbersome process, involving different possibilities and restrictions in clinical settings, as

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

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

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

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

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

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

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

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

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

163 The aims of the present study were: First, to explore adjustment across a wide range of  
164 domains. Among all identified domains that were mentioned above, measures of quality of  
165 life and self-concept were not available in the present study. However, all other aspects of  
166 psychological adjustment were represented and categorised into five main domains: cognitive,  
167 behavioural, emotional and social functioning, and satisfaction with appearance. The effects  
168 of gender, cleft visibility and the presence of an additional condition were evaluated, in  
169 addition to possible interactions for each of the five domains. Second, to identify a high risk  
170 group within each domain, in order to investigate whether risk factors co-varied across  
171 groups, or whether risk was restricted to specific domains of adjustment. Third, to present  
172 and discuss psychometric properties in relation to each outcome variable.

173 To the authors' knowledge, this is one of the first papers to include such a wide range of  
174 domains across a large sample, and to explore both risk and protective factors within a single  
175 study.

176 *Method*

177 *Setting*

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

## Participants

185

### 186 *Children*

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

192 In the cleft sample, 336 children were female and 509 were male. Children's cleft type  
193 included cleft lip and palate, CLP ( $n = 368$ ), cleft lip or cleft lip alveolus, CLA ( $n = 120$ )<sup>1</sup>,  
194 cleft palate, CP ( $n = 275$ ) or submucous cleft palate, SMCP ( $n = 59$ ). Information about the  
195 child's cleft type was missing for three children. For the purpose of the statistical analyses, the  
196 children were categorized into two groups: children with visible clefts (CLP and CLA,  $n =$   
197 488) and children with non-visible clefts (CP/SMCP,  $n = 354$ ). Among the girls, 51.8% had a  
198 non-visible cleft and 48.2% had a visible cleft. Among the boys, 31.4% had a non-visible  
199 cleft and 68.6% had a visible cleft. Some of the children were of non-Caucasian origin ( $n =$   
200 86/812, 10.6%), some of them adopted ( $n = 55/798$ ; 6.9% of the total sample).

### 201 *Parents*

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

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<sup>1</sup> 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.



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

### 209 *Additional conditions*

210 Information about additional conditions was found in the child's case records, discussed  
211 during the 10-year-old assessment, and/or was given by the child's parents. A total of 278  
212 children (33.3%) had one or several additional conditions, such as developmental delay  
213 (13.4%;  $n = 114$ ), learning difficulties (7.3%;  $n = 62$ ), dyslexia (5.5%;  $n = 47$ ), autism  
214 spectrum disorders (1.9%;  $n = 16$ ) and AD/HD (8.0%;  $n = 68$ ). Furthermore, some children  
215 had a diagnosed syndrome (9.3%;  $n = 79/847$ ), such as 22Q11.2, Treacher Collins, Goldenhar  
216 and Sticklers, with or without associated psychological and/or cognitive difficulties. While  
217 135 of the children had one extra diagnosed condition in addition to the cleft (16.1% of the  
218 total sample; 48.4% of the children with an additional condition), 79 of the children had two  
219 additional diagnoses (9.4%; 28.3%), while the remaining 65 had three or more conditions in  
220 addition to the cleft (7.7%; 23.3%).

### 221 Measures

#### 222 *Personality Inventory for Children (PIC)*

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

232 IQ on the Wechsler scales (Wirt et al., 1984). A Norwegian version of the instrument was  
233 used (Troland, 1988). Internal consistency ( $\alpha = .59-.86$ ;  $M = .74$ ), test-retest reliability ( $r =$   
234  $.46-.94$ ;  $M = .86$ ), and validity have been extensively evaluated and found to be satisfactory  
235 (Wirt et al., 1984).

#### 236 *Child Experience Questionnaire (CEQ)*

237 The CEQ (Pertschuk & Whitaker, 1982) reflects the child's self-reporting of social  
238 experiences on a 5-point Likert scale. The questions in the scale relate to topics such as  
239 relationships with friends ("I play with friends at school"), social isolation ("I try to hide from  
240 people"), and involvement in new experiences ("I meet new people"). Both positively and  
241 negatively worded items are included, to avoid systematic response bias. Scores are  
242 converted to a positive value so that high scores on the CEQ reflect positive social  
243 experiences. A mean total score was calculated. The scale has been shown to possess  
244 satisfactory internal consistency and a coherent factor structure (Emerson et al., 2004).

#### 245 *Strength and Difficulties Questionnaire (SDQ)*

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

257 been extensively validated, and cut-off scores presented by Goodman ([www.sdq.info](http://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  $2 \times 2 \times 2$  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 ( $\eta^2$ ) were calculated. Cohen's guidelines  
279 (1988) were used to interpret  $\eta^2$ : small effect: 0.01; medium effect: 0.059;  
280 large effect: 0.138. Effect sizes were only calculated in cases of statistical

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

283 ii. Comparisons between the cleft sample and reference groups/norms and/or  
284 clinical cut-off scores are given. Reference groups for the SDQ were large  
285 national same-aged and non-cleft samples (Self-reports: Van Roy et al.,  
286 2006; Parent reports: Van Roy et al., 2010), which were compared to  
287 children with a cleft and no additional condition. Independent sample *t*-tests  
288 provided Mean scores (M) and Standard deviations (SD) which could be  
289 directly compared with scores from the reference group. Calculations of  
290 effect size were performed using Cohen's *d* in cases of significant  
291 differences (Cohen, 1988; 0.2 = small, 0.5 = medium, and 0.8 = large effect).

292 iii. Identification of a high risk group according to norms (PIC: clinical cut-off  
293 scores indicating moderate or severe problems) or according to scores below  
294 the 10<sup>th</sup> percentile (SWA and CEQ) or above the 90<sup>th</sup> percentile (SDQ). Cut-  
295 off scores from large national samples were used for the SDQ. A  
296 dichotomous variable was created in order to explore the characteristics of  
297 the high risk groups with respect to gender, cleft visibility, and the presence  
298 or absence of an additional condition. Chi-square analyses were used when  
299 investigating differences between the categorical variables.

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

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

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

## 316 Results

### 317 *General Adjustment*

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

### 320 *Adjustment (PIC)*

321  $2 \times 2 \times 2$  ANOVA: There were no interactions, while two main effects were found, related to  
322 cleft visibility and the presence of an additional condition (Table 1). Children with a CP (with  
323 and without an additional condition) had significantly less adjustment problems (EMM =  
324 55.9, SE = .84) than the total sample of children with CLP (EMM = 58.4, SE = .86;  $F(1,435)$   
325 = 4.12,  $p < .05$ ). However, calculations of effect size showed that this effect was small ( $\eta^2 =$   
326 0.007). A main effect was also found between children with a cleft only (EMM = 50.6, SE =  
327 .64) and children with a cleft and an additional condition (Cleft +: EMM = 63.7, SE = 1.02;  $F$   
328 (1,435) = 116.95,  $p < .001$ ), with a very large effect size ( $\eta^2 = 0.205$ ).

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

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

337 *Total difficulties score (SDQ)*

338  $2 \times 2 \times 2$  ANOVA: As can be seen in Table 1, only one main effect was found on self- and  
339 parent reports, highlighting the risk of more psychological difficulties in children with an  
340 additional condition (EMM = 13.1, SE = .51) when compared to children with a cleft only  
341 (EMM = 9.5, SE = .42;  $F(1, 288) = 29.75$ ,  $p < .001$ ;  $\eta^2 = 0.092$ ). The same effect was found  
342 in parent reports (Cleft only: EMM = 5.8, SE = .45; Cleft +: EMM = 11.5, SE = .53;  $F(1, 294)$   
343  $= 67.90$ ,  $p < .001$ ). Effect size was large ( $\eta^2 = 0.182$ ).

344 *Reference group comparisons:* On self-reports, girls with a cleft without an additional  
345 condition had similar scores ( $M = 9.8$ ,  $SD = 4.9$ ) to girls from the reference group ( $M = 10.1$ ,  
346  $SD = 5.1$ ;  $t(1431) = 0.51$ ,  $p > .05$ ). The same was found in parent reports (Cleft:  $M = 6.1$ ,  $SD$   
347  $= 4.8$ ; Ref.gr.:  $M = 5.7$ ,  $SD = 4.8$ ;  $t(4121) = 0.56$ ,  $p > .05$ ). Boys with a cleft and no  
348 additional condition had less psychological adjustment problems on both self- ( $M = 9.2$ , SE =  
349 4.9) and parent reports ( $M = 5.5$ ,  $SD = 4.4$ ) than boys from the reference group (Self-reports:  
350  $M = 10.3$ ,  $SD = 5.2$ ;  $t(1560) = 2.10$ ,  $p < .05$ ;  $d = -.22$ ; Parent reports:  $M = 6.6$ ,  $SD = 5.2$ ;  $t$   
351  $(4180) = 2.15$ ,  $p < .05$ ;  $d = -.23$ ).

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

#### 360 *Cognitive Function*

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

#### 365 *Intellectual Screening (PIC)*

366  $2 \times 2 \times 2$  ANOVA: There were no interactions and two main effects (Table 1). As could be  
367 expected, children with an additional condition had higher scores on the Intellectual Screening  
368 scale,  $F(1, 436) = 268.27$ ,  $p < .001$ , indicating more cognitive problems (EMM = 86.0, SE =  
369 1.57) than children with a cleft only (EMM = 55.7, SE = .98). Effect size was very large ( $\eta^2 =$   
370 0.360). The second significant main effect was that children with a CP had more cognitive  
371 problems (EMM = 72.8, SE = 1.29) than children with a visible cleft (EMM = 68.9, SE =  
372 1.32;  $F(1, 436) = 4.47$ ,  $p < .05$ ;  $\eta^2 = 0.006$ ).

373 *Cut-off scores:* The cut-off score indicating elevations that are clinically significant are set at  
374  $> 59T$  for the Intellectual Screening subscale. Hence, mean scores were above the clinical  
375 range for boys and girls, and irrespective of cleft visibility, when analyses were performed

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

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

#### 386 *Hyperactivity (PIC)*

387  $2 \times 2 \times 2$  ANOVA: As can be seen in Table 1, there were two significant 2-way interactions,  
388 one between gender and an additional condition ( $F(1,435) = 4.35, p < .05$ ), the other one  
389 between cleft visibility and an additional condition ( $F(1,435) = 3.91, p < .05$ ). The patterns  
390 of these interactions were that the impact of an additional condition on problems with  
391 hyperactivity seemed to be stronger for the girls than for the boys, while the opposite pattern  
392 was the case in children without an additional condition. In addition, the impact of an  
393 additional condition was stronger in children with CLP than in children with CP. Effect sizes  
394 were small for both interactions ( $\eta^2 < 0.010$ ), hence the details of the ANOVA are not  
395 reported in further detail.

396 There were two main effects. As could be expected, children with an additional condition had  
397 higher scores (EMM = 53.2, SE = 1.00) than children with a cleft only (EMM = 45.8, SE =  
398 .63;  $F(1, 436) = 38.76, p < .001$ ;  $\eta^2 = 0.360$ ) on the Hyperactivity scale. The second main  
399 effect indicated that children with CLP had more problems with hyperactivity (EMM = 51.0,



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

403 *Cut-off scores:* The cut-off score indicating elevations that are clinically significant are set at  
404 > 59T for the Hyperactivity subscale, meaning that although statistics indicated significant  
405 differences between subgroups, mean scores were still within the normal range for all groups.

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

#### 412 *Attention and Hyperactivity (SDQ)*

413  $2 \times 2 \times 2$  ANOVA: There were no interactions and one main effect on self-reports, while parent  
414 reports pointed to two main effects (Table 1). Children with a cleft and an additional  
415 condition expectedly had more problems with attention and/or hyperactivity (Self-reports:  
416 EMM = 5.0, SE = .21; Parent reports: EMM = 4.6, SE = .24) than children with a cleft only  
417 (Self-reports: EMM = 3.7, SE = .17;  $F(1, 288) = 21.27, p < .001; \eta^2 = 0.075$ ; Parent reports:  
418 EMM = 2.3, SE = .20;  $F(1, 294) = 55.56, p < .001; \eta^2 = 0.157$ ). The second main effect was  
419 found in parent reports only: boys had more problems with attention and/or hyperactivity  
420 (EMM = 3.8, SE = .21) than girls (EMM = 3.1, SE = .23;  $F(1, 288) = 4.77, p < .05$ ). Effect  
421 size, however, was small ( $\eta^2 = 0.013$ ).

422 *Reference group comparisons:* Girls with a cleft and no additional condition ( $M = 3.4, SD =$   
423  $1.98$ ) had similar scores as girls from the reference group on self-reports ( $M = 3.5, SD = 2.0; t$

424 (1431) = 0.44,  $p > .05$ ) and on parent reports (Cleft:  $M = 2.3$ ,  $SD = 2.3$ ; Ref.gr.:  $M = 2.2$ ,  $SD$   
 425 = 2.0;  $t(4154) = 0.44$ ,  $p > .05$ ). The same was the case for the boys on self-reports ( $M = 4.0$ ,  
 426  $SD = 2.1$ ), as compared to those from the reference group ( $M = 3.8$ ,  $SD = 2.1$ ;  $t(1561) = 0.95$ ,  
 427  $p > .05$ ). The parents of boys with a cleft, on the other hand, reported significantly less  
 428 problems with attention and hyperactivity ( $M = 2.5$ ,  $SD = 2.07$ ) than parents from the  
 429 reference group ( $M = 3.0$ ,  $SD = 2.4$ ;  $t(4180) = 2.12$ ,  $p < .05$ ;  $d = -.22$ ).

430 *High risk group analysis:* Cut-off scores identified 44 children (15.1%) at high risk for  
 431 hyperactivity problems on self-reports, and 43 children (14.4%) according to parent reports.  
 432 There were no gender differences (Self-reports:  $\chi^2 = 1.42$ ,  $p > .05$ ; Parent reports:  $\chi^2 = 1.03$ ,  $p$   
 433  $> .05$ ), nor differences related to cleft visibility (Self-reports:  $\chi^2 = 2.32$ ,  $p > .05$ ; Parent  
 434 reports:  $\chi^2 = .22$ ,  $p > .05$ ). As expected, there were significantly more children with a cleft  
 435 and an additional condition in the high risk group (Self-reports: 24.1%,  $n = 26$ ; Parent reports:  
 436 27.8%,  $n = 32$ ) compared to children with cleft only (Self-reports: 9.8%,  $n = 18$ ,  $\chi^2 = 10.73$ ,  $p$   
 437  $< .01$ ; Parent reports: 6.0%,  $n = 11$ ;  $\chi^2 = 27.22$ ,  $p < .001$ ).

#### 438 *Behavioural conduct*

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

#### 441 *Withdrawal (PIC)*

442  $2 \times 2 \times 2$  ANOVA: Analyses revealed no interactions and one main effect (Table 1). Children  
 443 with an additional condition had higher scores on the Withdrawal scale ( $EMM = 54.2$ ,  $SE =$   
 444  $.89$ ) than in cases of a cleft only ( $EMM = 51.1$ ,  $SE = .55$ ;  $F(1, 436) = 8.92$ ,  $p < .01$ ). Effect  
 445 size was small ( $\eta^2 = 0.020$ ).

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

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

452 *Conduct problems (SDQ)*

453  $2 \times 2 \times 2$  ANOVA: There was one main effect (Table 1). Children with a cleft and an additional  
454 condition had more conduct problems (Self-reports: EMM = 1.9, SE = .15; Parent reports:  
455 EMM = 1.7, SE = .14) than children with a cleft only (Self-reports: EMM = 1.5, SE = .12;  
456  $F(1, 288) = 5.30, p < .05; \eta^2 = 0.017$ ; Parent reports: EMM = 1.0, SE = .12;  $F(1, 295) = 13.78,$   
457  $p < .001; \eta^2 = 0.044$ ).

458 *Reference group comparisons:* Girls with a cleft and no additional condition had similar  
459 scores as girls from the reference group on self-reports (Cleft: M = 1.5, SD = 1.35; Ref.gr.: M  
460 = 1.4, SD = 1.31;  $t(1431) = 0.66, p > .05$ ) and parent reports (Cleft: M = 1.0, SD = 1.19;  
461 Ref.gr.: M = 1.1, SD = 1.4;  $t(4154) = 0.63, p > .05$ ). The same was the case for boys on  
462 parent reports (Cleft: M = 1.1, SD = 1.29; Ref.gr.: M = 1.0, SD = 1.2;  $t(4180) = 0.84, p >$   
463  $.05$ ). On self-reports, boys with a cleft reported significantly less conduct problems (M = 1.5,  
464 SD = 1.48) than the reference group (M = 2.0, SD = 1.74;  $t(1561) = 3.36, p < .001; d = -.31$ ).

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

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

472 *Social experiences*

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

474 *Child Experience Questionnaire (CEQ)*

475  $2 \times 2 \times 2$  ANOVA: There were no interactions and only one main effect (Table 1): children with  
476 a cleft and an additional condition reported less positive social experiences (EMM = 2.4, SE =  
477 .03) than children with a cleft only (EMM = 2.6, SE = .02;  $F(1, 592) = 26.99, p < .001; \eta^2 =$   
478 0.043).

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

482 *High risk group analysis*: Percentile analyses revealed that a mean of 2.10 or lower was  
483 indicative of high psychosocial risk (< 10<sup>th</sup> percentile). The high risk group consisted of 70  
484 children (11.8%). The presence of an additional condition was the only significant risk factor  
485 (8.3%,  $n = 34$  vs. 19.5%,  $n = 36$ ;  $\chi^2 = 15.22, p < .001$ ). There were no gender differences ( $\chi^2$   
486 = 1.02,  $p > .05$ ), and no differences related to cleft visibility ( $\chi^2 = .28, p > .05$ ).

487 *Peer problems (SDQ)*

488  $2 \times 2 \times 2$  ANOVA: There were no interactions and only one main effect on self-reports and  
489 parent reports (Table 1). Children with a cleft and an additional condition reported more peer  
490 problems (Self-reports: EMM = 2.6, SE = .17; Parent reports: EMM = 2.5, SE = .17) than  
491 children with a cleft only (Self-reports: EMM = 1.8, SE = .14;  $F(1, 288) = 11.13, p < .01$ ;

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

494 *Reference group comparisons:* Compared to reference groups, girls with a cleft and no  
 495 additional condition reported the same level of peer problems ( $M = 1.9$ ,  $SD = 1.7$ ) as girls  
 496 from the reference group on self-reports ( $M = 1.9$ ,  $SD = 1.7$ ;  $t(1431) = 0.00$ ,  $p > .05$ ) and  
 497 parent reports (Both groups:  $M = 1.1$ ,  $SD = 1.6$ ;  $t(4154) = 0.00$ ,  $p > .05$ ). Boys with a cleft  
 498 reported significantly less peer problems ( $M = 1.7$ ,  $SD = 1.5$ ) than the reference group on self-  
 499 reports ( $M = 2.1$ ,  $SD = 1.8$ ;  $t(1561) = 2.23$ ,  $p < .05$ ;  $d = -.24$ ) and on parent reports (Cleft:  $M$   
 500  $= .8$ ,  $SD = 1.3$ ; Ref.gr.:  $M = 1.3$ ,  $SD = 1.7$ ;  $t(4180) = 2.99$ ,  $p < .001$ ;  $d = -.33$ ).

501 *High risk group analysis:* Cut-off scores identified 34 children (11.7%) at high risk for peer  
 502 problems according to self-reports and 47 children (15.8%) according to parent reports. There  
 503 were no gender differences (Self-reports:  $\chi^2 = .05$ ,  $p > .05$ ; Parent reports:  $\chi^2 = .05$ ,  $p > .05$ ),  
 504 and no difference related to cleft visibility ( $\chi^2 = .01$ ,  $p > .05$ ;  $\chi^2 = 1.67$ ,  $p > .05$ ). There were  
 505 more children with an additional condition in the high risk group (Self-reports: 17.6%,  $n = 19$ ;  
 506 Parent reports: 31.3%,  $n = 36$ ) than in cases of a cleft only (Self-reports: 8.2%,  $n = 15$ ;  $\chi^2 =$   
 507  $5.81$ ,  $p < .05$ ; Parent reports: 6.0%,  $n = 11$ ;  $\chi^2 = 34.01$ ,  $p < .001$ ).

#### 508 *Emotional Adjustment*

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

#### 511 *Depressive Symptoms and Anxiety (PIC)*

512  $2 \times 2 \times 2$  ANOVA: Analyses revealed no interactions and only one main effect (Table 1).

513 Children with a cleft and an additional condition had more problems with depression ( $EMM =$   
 514  $58.0$ ,  $SE = 1.10$ ) than children with a cleft only ( $EMM = 50.4$ ,  $SE = .69$ ;  $F(1, 435) = 34.64$ ,  $p$   
 515  $< .001$ ). The same was the case for anxiety symptoms (Cleft +:  $EMM = 59.2$ ,  $SE = 1.07$ ;

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

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

521 *High risk group analysis:* There were 15 children (3.5%) at high risk for depression and 10  
522 (2.3%) at high risk for anxiety-related conditions. There were no differences related to cleft  
523 visibility ( $\chi^2 = .49$  and  $.59$ ,  $p > .05$ , respectively), and no gender differences ( $\chi^2 = 2.30$  and  
524  $.55$ ,  $p > .05$ ) in the high risk group. There were significantly more children with an additional  
525 condition (10.3%,  $n = 12$  and 5.7%,  $n = 7$ ) than children with a cleft only (1.0%,  $n = 3$ ;  $\chi^2 =$   
526  $21.85$ ,  $p < .001$  and 1.0%,  $n = 3$ ;  $\chi^2 = 8.97$ ,  $p < .01$ ).

527 *Emotional difficulties (SDQ)*

528  $2 \times 2 \times 2$  ANOVA: There was one interaction in self-and parent reports, two main effects in  
529 self-reports, and one main effect in parent reports (Table 1). On self-reports, the pattern of the  
530 interaction was that while the girls with a cleft had rather high scores whether they had an  
531 additional condition or not, the impact of an additional condition seemed more important in  
532 boys ( $F(1, 288) = 3.95$ ,  $p < .05$ ). In parent reports, the interaction was related to gender and  
533 cleft visibility ( $F(1, 288) = 8.80$ ,  $p < .01$ ). Girls with a visible cleft reported less emotional  
534 difficulties than girls with a non-visible cleft, while the opposite was the case for boys.  
535 However, effect sizes were small for both interactions ( $\eta^2 < 0.017$ ).

536 The main effects in self-reports involved gender and the presence of an additional condition.  
537 Girls reported more emotional difficulties ( $EMM = 3.4$ ,  $SE = .22$ ) than boys ( $EMM = 2.8$ ,  $SE$   
538  $= .19$ ;  $F(1, 288) = 4.35$ ,  $p < .05$ ). Effect size, however, was small ( $\eta^2 = 0.013$ ). The other  
539 main effect was once again related to the presence of an additional condition (Cleft:  $EMM =$

540 2.5, SE = .19; Cleft+: EMM = 3.7, SE = .23;  $F(1,288) = 16.35, p < .001$ ). There was only  
 541 one main effect in parent reports, associated with the presence of an additional condition ( $F$   
 542  $(1,295) = 23.96, p < .001$ ). Effect sizes were within the medium range on self-reports ( $\eta^2 =$   
 543  $0.046$ ) and parent reports ( $\eta^2 = 0.069$ ).

544 *Reference group comparisons:* Girls with a cleft and no additional condition ( $M = 3.0, SD =$   
 545  $2.2$ ) reported similar levels of emotional problems as girls from the reference group on self-  
 546 reports ( $M = 3.0, SD = 2.2; t(1431) = 0.00, p > .05$ ), and had more emotional problems  
 547 according to parent reports (Cleft:  $M = 1.8, SD = 1.9$ ; Ref.gr.:  $M = 1.4, SD = 1.8; t(4154) =$   
 548  $1.95, p = .051; d = .22$ ). However, this difference was not statistically significant. Boys with  
 549 a cleft ( $M = 2.2, SD = 2.1$ ) reported similar levels of emotional difficulties as the reference  
 550 group on self-reports ( $M = 2.2, SD = 2.1; t(1561) = 0.00, p > .05$ ) and parent reports (Cleft:  
 551  $M = 1.2, SD = 1.4$ ; Ref.gr.:  $M = 1.2, SD = 1.7; t(4180) = 0.00, p > .05$ ).

552 *High risk group analysis:* There were 43 children (14.8%) at high risk for emotional problems  
 553 according to self-reports, and 38 children (12.8%) according to parent reports. Self-reports  
 554 revealed more girls (20.3%,  $n = 25$ ) than boys (10.7%,  $n = 18$ ) in the high risk group ( $\chi^2 =$   
 555  $5.21, p < .05$ ), while this difference was not significant in the parent reports ( $\chi^2 = .53, p > .05$ ).  
 556 Self-reports also identified more children with a CP (21.0%,  $n = 22$ ) in the high risk group  
 557 than children with CLP (11.4%,  $n = 21; \chi^2 = 4.89, p < .05$ ), while parent reports did not ( $\chi^2 =$   
 558  $2.28, p > .05$ ). While 10.4% ( $n = 19$ ) of the children with a cleft only were found in the high  
 559 risk group, this was the case for 22.2% ( $n = 24$ ) of the children with an additional condition  
 560 ( $\chi^2 = 7.56, p < .01$ ). Approximately the same pattern was found in parent reports (5.5%,  $n =$   
 561  $10$  vs. 24.3%,  $n = 28; \chi^2 = 22.63, p < .001$ ).

562

#### *Satisfaction with appearance*

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

565  $2 \times 2 \times 2$  ANOVA: As can be seen in Table 1, analyses revealed only one main effect, children  
566 with an additional condition reporting less satisfaction with appearance (EMM = 8.1, SE =  
567 .11) than children with a cleft only (EMM = 8.5, SE = .07;  $F(1,676) = 9.23, p < .01$ ).  
568 However, effect size was small ( $\eta^2 = 0.014$ ).

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

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

583 *High risk group analysis:* Percentile analyses revealed that a mean of 6.18 or lower was  
584 indicative of high risk for dissatisfaction with total appearance ( $< 10^{\text{th}}$  percentile). A total of  
585 66 children (9.7%) were found within the high risk group. There were no gender differences  
586 ( $\chi^2 = .74, p > .05$ ), no differences related to visibility of cleft ( $\chi^2 = .44, p > .05$ ), and no



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

589 *Risk groups across measures*

590 In order to compare risk groups across measures, five new variables were created<sup>2</sup>. These five  
591 variables recorded the children that had been identified as being at high risk of cognitive,  
592 behavioural, social and/or emotional problems, and/or at high risk for dissatisfaction with  
593 appearance, irrespective of which outcome measure that had been used initially. In addition,  
594 children reporting scores within the borderline range were identified and recorded. Hence, as  
595 an example, children at risk for depressive symptoms and anxiety (PIC), and/or those  
596 identified at risk for emotional difficulties (SDQ) were recorded in the new variable named  
597 “Emotional adjustment”. An overview of the frequency of children with a cleft within the  
598 normal range, or in the borderline and high risk groups according to the five new variables is  
599 presented in Table 2.

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

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

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<sup>2</sup> 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.

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

612 Correlations between the five risk groups were calculated. Most correlations were significant,  
613 and varied from no associations to moderate associations. The strongest association was  
614 found between social and emotional risk ( $r = .38, n = 598, p < .001$ ). The other correlations  
615 were, in order of strength of association: emotional and behavioural risk ( $r = .35, n = 708, p <$   
616  $.001$ ), emotional and cognitive risk ( $r = .31, n = 711, p < .001$ ), social and behavioural risk ( $r$   
617  $= .28, n = 596, p < .001$ ), behavioural and cognitive risk ( $r = .28, n = 708, p < .001$ ) and  
618 cognitive and social risk ( $r = .23, n = 598, p < .001$ ). The remaining four correlations were  
619 weak or non-significant: appearance and social risk ( $r = .18, n = 572, p < .001$ ), appearance  
620 and cognitive risk ( $r = .12, n = 631, p < .01$ ), appearance and emotional risk ( $r = .07, n = 620,$   
621  $p > .05$ ) and appearance and behavioural risk ( $r = 0.03, n = 633, p > .05$ ).

## 622 *Psychometric properties*

### 623 *Correlations across measures and informants*

624 Calculations of convergent validity and levels of agreement between child and parent reports  
625 are presented in Table 3. Correlations between the CEQ and the Peer problems subscale of  
626 the SDQ were moderate, as was the case for levels of agreement between child and parent  
627 reports for the SDQ. Correlations were similar or higher than previously reported (Goodman,  
628 2001; Van Roy et al, 2010).

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

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

636 *Internal consistency*

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

644 Discussion

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

650 The prevalence of cognitive, behavioural, emotional, social, and appearance-related risk was  
651 significantly associated with the presence of an additional condition in all measures, while the  
652 effect of cleft visibility and gender seemed to be less important at age 10. Approximately  
653 60% of the children were not at high risk in any of the adjustment domains. Less than 25%  
654 were at high risk in one domain only, while approximately 15% were at high risk in two or  
655 more domains of adjustment.

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

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

661 *Psychological functioning: The role of an additional condition*

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

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

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

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

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

708 *Risk and Protection*

709 Boys with a cleft only showed more positive adjustment on several domains compared to  
710 same-aged boys from the reference groups, while girls with a cleft had similar scores as girls  
711 from the general population. In addition, almost 60% of the children within the sample had  
712 scores within the normal/borderline range on all domains of adjustment. This could indicate  
713 the presence of protective factors that counteract the consequences of potential risk. The  
714 results suggest that most children with a cleft cope well, in spite of specific challenges that are  
715 known to be associated with living with a visible difference. Further, the lack of strong  
716 associations between the risk groups suggest that risk seem to be domain-specific, and not  
717 general in children with a cleft. This could indicate that interventions tailored within specific  
718 domains of risk may be efficient for most children with this condition. Of the five domains,  
719 only social and emotional risk and emotional and behavioural risk were found to be associated  
720 at a level that was considered clinically significant. However, the magnitude of these  
721 associations was moderate. Additionally, being dissatisfied with subjective appearance at age  
722 10 was not associated with emotional, behavioural or psychosocial difficulties. Interestingly,  
723 a similar finding was reported in adults with a cleft (Roberts and Mathias, 2012), while other  
724 studies have pointed to the importance of subjective appearance evaluations for psychological  
725 adjustment in older participants (Feragen et al., 2010; Mani et al., 2013). For the present age  
726 group, the findings could hence point towards the effectiveness of interventions which taps  
727 into specific domains of risk, such as social skills training, cognitive-behavioural  
728 interventions, or interventions directed towards reducing emotional distress (Robinson et al.,  
729 1996; Maddern and Owen, 2004; Kapp-Simon et al., 2005; Bessell et al., 2012), when  
730 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)

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

759 Social disadvantage due to the visible difference have been reported previously (Murray et al.,  
760 2010), and has been supported by neuropsychological findings related to social function  
761 (Canady et al., 2007). However, the current findings did not indicate that the children in this  
762 study were at social and emotional risk because of cleft visibility. Such findings address the  
763 need for research to identify other risk factors in this population, and to acknowledge positive  
764 adjustment factors (Egan et al., 2011; Roberts and Mathias, 2012), in order to capture the  
765 complexity of adjustment to a visible difference (Stock et al., 2013). Longitudinal studies are  
766 ultimately needed in order to address the directionality of associations, and whether risk  
767 groups would be found within the same adjustment domains in later developmental stages.

#### 768 *Gender differences*

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

774 Within the cleft sample, boys were more at risk for problems with attention than girls  
775 according to parent reports, while girls were at greater emotional risk on self-reports. Such  
776 gender differences are in line with findings from the general population (Rønning et al., 2004;  
777 Van Roy et al., 2006; Van Roy et al., 2010). However, when comparing the cleft sample with  
778 the reference group, parent-reports indicated that boys with a cleft had less attention problems  
779 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

804 been shown to increase from childhood to adolescence, especially in girls (Dekker et al.,  
805 2007; Smolak, 2012; Snyder and Pope, 2010), results from samples with wide age ranges may  
806 be imprecise and gender differences be blurred by differences related to age. Clearly defined  
807 age groups are needed to explore adjustment across different developmental stages. Gender  
808 differences in the general population point to the importance of gender-specific results also in  
809 the cleft literature. In order to be able to explore this, large samples are needed; a factor that  
810 probably explains the choices related to age and gender made in many studies.

811 *Generic vs. specific measures in cleft clinics*

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

818 The fact that cleft visibility did not affect the outcome measures could be explained in  
819 different ways. One interpretation is that cleft visibility in itself is not the main issue for  
820 psychological adjustment, as has been demonstrated by several recent studies (Moss, 2005;  
821 Appearance Research Collaboration, 2009; Feragen et al., 2010). Another interpretation  
822 could be that generic measures are not sensitive or specific enough to actually highlight  
823 existing difficulties or condition-specific challenges. A third interpretation could be that  
824 children with a cleft, in spite of, or because of the challenges involved in their condition, still  
825 develop an ability to cope with their condition, resulting in positive adjustment. The lack of  
826 strong associations between the different risk groups, and the positive adjustment findings in  
827 comparison to reference groups, could support this final hypothesis. Further, the lack of  
828 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

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

889 The SDQ is user-friendly and quick to administer, is widely available and free to use, and has  
890 been translated into several languages. Norms have been provided for many different  
891 countries, and reference groups are also available as a consequence of the number of studies  
892 using it. Unfortunately, internal reliability in this and in other studies (Goodman, 2001;  
893 Rønning et al., 2004; Stone et al., 2010) has been shown to be poor, unacceptable or  
894 questionable for some subscales, such as the ones measuring conduct difficulties, peer  
895 problems and emotional difficulties. The subscale measuring problems with attention and/or  
896 hyperactivity had good reliability on parent reports, while self-reports at age 10 were  
897 questionable. Nonetheless, the Total difficulties score showed good reliability and correlated  
898 highly with the general adjustment scale from the PIC. It is already used in some countries  
899 which have centralised cleft lip and palate treatment, which would make comparisons across  
900 countries possible and valuable in the future.

901 The CEQ has been used in cleft research previously, but published norms are not available.  
902 Unfortunately, the measure has been used differently across studies and results are sometimes  
903 calculated in alternate ways, making meaningful comparisons more challenging. Although

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

908 The SWA has also been used in a number of cleft studies and was the only measure in the  
909 present study which seemed to point to challenges related to cleft visibility. The measure is  
910 easy to administer and interpret, and demonstrated excellent internal reliability within the  
911 current sample. The SWA appears to be a useful measure, but again, unfortunately no  
912 published norms are available at present, and convergent validity could not be computed since  
913 other appearance-related measures were not used in the present data-set. Normative data have  
914 been reported to exist for a UK sample, and are reported in Berger and Dalton (2009).  
915 However, the age range includes children and adolescents, complicating comparisons, and  
916 most probably obscuring age-specific differences in satisfaction with appearance.

#### 917 *Strengths and Limitations*

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

929 running a 2×2×2 ANOVA instead of several *t*-tests, the chances of Type I error were kept at  
930 5%, and estimated marginal means were adjusted for the other variables in the model. Hence,  
931 more correct estimations of the variable's effect on outcome were provided.

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

937 Due to its retrospective nature, the study was restricted by the measures that had been used  
938 during routine assessments. Hence, even if most areas of research were addressed that had  
939 been identified in recent systematic reviews and book chapters, some measures may not have  
940 been optimal in capturing specific issues of adjustment. As an example, cognitive risk may  
941 have been better assessed with tests of cognitive performance and abilities. Another  
942 limitation could be the lack of data for the children with severe developmental problems who  
943 did not go through the routine assessment, since they were not able to complete any of the  
944 measures used. Their presence in the sample would probably have impacted on the mean  
945 scores for most variables, increasing the findings related to risk in the group of children with  
946 an additional condition, and needs to be acknowledged. Further, adjustment to a visible  
947 difference involves a combination of psychological and societal factors that were not  
948 accounted for in the present study, such as individual characteristics, cognitive processes  
949 (such as attribution style or coping strategies), family factors and social support, in addition to  
950 socio-cultural factors. An additional variable of potential importance in children with a cleft  
951 is related to problems with speech, a variable which has been shown to be associated with  
952 social difficulties (Watterson et al., 2013). Unfortunately, speech outcomes other than the  
953 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.

<b>Psychological adjustment</b>		<b>Main effects and Interactions</b>	<b>F</b>	<b>R<sup>2</sup></b>
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
<b>Cognitive function</b>				
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
<b>Behavioural conduct</b>				
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
<b>Social experiences</b>				
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
<b>Emotional adjustment</b>				
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**	
<b>Appearance satisfaction</b>				
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

		<b>Normal range</b>	<b>Borderline</b>	<b>High risk</b>
	<b><i>n</i></b>	<b>% (<i>n</i>)</b>	<b>% (<i>n</i>)</b>	<b>% (<i>n</i>)</b>
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			<i>n</i>	Pearson's <i>r</i>
		S-S	S-P	P-P		
<b>General adjustment</b>						
Total difficulties score (SDQ)	Total difficulties score (SDQ)		X		281	.45***
<b>Cognitive function</b>						
Intellectual Screening (PIC)	Hyperactivity (PIC)			X	436	.29***
Attention problems (SDQ)	Attention problems (SDQ)		X		281	.42***
<b>Behavioural difficulties</b>						
Conduct problems (SDQ)	Conduct problems (SDQ)		X		282	.32***
<b>Social experiences</b>						
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***
<b>Emotional adjustment</b>						
Depressive symptoms (PIC)	Anxiety (PIC)			X	436	.84***
Emotional problems (SDQ)	Emotional problems (SDQ)		X		282	.28***
<b>Satisfaction with appearance</b>						
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	<i>n</i>	<i>α</i>		
<b>Personality Inventory for Children</b>	PIC	Adjustment	437	.81	Good	
		Intellectual Screening		.61	Questionable	
		Withdrawal		.57	Poor	
		Depression		.83	Good	
		Anxiety		.75	Acceptable	
		Hyperactivity		.48	Unacceptable	
<b>Child Experience Questionnaire</b>	CEQ		550	.73	Acceptable	
<b>Strengths and Difficulties Questionnaire</b>	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	
<b>Satisfaction with Appearance scales</b>	SWA		632	.89	Good/Excellent	