1

2

3

4

5

Speech, language and reading in 10-year-olds with cleft: Associations with teasing, satisfaction with speech and psychological adjustment

8

9

10

11

12

13

14

15

16

17

18

19

20

21
22
23
24
25

Abstract

Background: Despite the utilisation of multidisciplinary services, little research has addressed issues involved in the care of those with cleft lip and/or palate across disciplines. The aim was to investigate associations between speech, language, reading and reports of teasing, subjective satisfaction with speech and psychological adjustment.

Design: Cross-sectional data collected during routine, multidisciplinary assessments in a centralised treatment setting, including speech and language therapists and clinical psychologists.

Participants: Children with cleft with palatal involvement aged 10 from three birth cohorts (*n* = 170) and their parents.

Outcome measures: *Speech*: SVANTE-N. *Language*: Language 6-16 (Sentence recall, Serial recall, Vocabulary, and Phonological awareness). *Reading*: Word Chain Test and Reading Comprehension Test. *Psychological measures*: Strengths and Difficulties Questionnaire and extracts from the Satisfaction with Appearance Scale and Child Experience Questionnaire.

Results: Reading skills were associated with self- and parent- reported psychological adjustment in the child. Subjective satisfaction with speech was associated with psychological adjustment, while not being consistently associated with speech therapists' assessments. Parent-reported teasing was found to be associated with lower levels of reading skills. Having a medical and/or psychological condition in addition to the cleft was found to impact significantly on speech, language and reading.

Conclusions: Cleft teams need to be aware of speech, language and/or reading problems as potential indicators of psychological risk in children with cleft. This study highlights the importance of multiple reports (self, parent and specialist) and a multidisciplinary approach to cleft care and research.

Key Words: Cleft lip and palate; speech; language; reading; psychological adjustment; teasing.

53 **Introduction**

54   The management of a child born with a cleft lip and/or palate (CL/P) can be complex,

55   involving a range of disciplines and interconnected treatment pathways. Following

56   the centralisation of cleft services in a number of European countries,

57   recommendations have been made to support the implementation of multidisciplinary

58   care (Sandy et al., 1998; Sandy et al., 2012). Many treatment centers worldwide

59   now follow these recommendations, involving surgeons, orthodontists, speech and

60   language therapists and psychologists, among others, within one team. This

61   diversity of team members can provide a foundation for more complex and complete

62   collaboration (Fox and Stone, 2013). Despite this, individuals working within

63   multidisciplinary teams often keep to their own independent scopes of practice (Fox

64   and Stone, 2013). Consequently, compared to the total number of studies, little

65   research has addressed issues involved in the care of those with CL/P across

66   disciplines.

67   One example of this pertains to associations between speech development,

68   language skills, reading ability and psychological variables. Several studies have

69   described potential problems related to the development of speech, language or

70   reading in children with CL/P (e.g. Kuehn and Moller, 2000; Richman and Ryan,

71   2003; Scherer et al., 2008; Hardin-Jones and Chapman, 2011), as well as in relation

72   to psychological, emotional and social adjustment (e.g. Turner et al., 1997; Hunt et

73   al., 2005; Rumsey and Stock, 2013), yet potential associations across the two

74   disciplines have received less attention.

75   A minority of studies in the field of CL/P have speculated on the possible overlap of

76   these two disciplines. Early research indicated a possible link between speech

77 difficulties and parent-reported behavioral problems in the child (McWilliams and

78 Musgrave, 1972). Simonds and Heimburger (1978) found that children with CL/P

79 and articulation difficulties were more likely to have psychiatric diagnoses, difficulties

80 with learning and problems related to psychological and interpersonal adjustment.

81 However, standardised measures have not been consistently used and findings have

82 not always been replicated (Richman, 1976). More recently, Millard and Richman

83 (2001) found an association between parent- and teacher-reported scores of

84 depression and anxiety and speech difficulties in children with nonsyndromic cleft

85 palate only (CP), although speech was not assessed by a speech and language

86 therapist.

87 Although only a few studies have investigated the direct impact of speech, language

88 and reading on psychological adjustment in children with cleft, some research has

89 examined the impact of neurobiological aspects. These studies have indicated that

90 abnormal brain structures in children with CL/P may influence cognitive function,

91 including language and reading, in addition to behavioural and speech outcomes

92 (Boes et al., 2007; Conrad et al., 2010; Nopoulos et al., 2010; Conrad et al., 2014).

93 Perceptions of teasing may be another important factor in the relationship between

94 speech and psychological adjustment in children with CL/P. Some early studies

95 suggested that difficulties with speech may invite negative reactions from others,

96 resulting in psychological distress and low self-esteem in the child (Richman, 1983;

97 Kapp-Simon et al., 1992). In self-reports, young people with CL/P have reported

98 teasing perceived by them as related to aspects of their speech (Turner et al., 1997;

99 Hunt et al., 2006; Noor and Musa, 2007; Havstam et al., 2011). More recently,

100 Watterson and colleagues (2013) demonstrated an association between perceived

101 speech problems and negative social acceptance, while subjective perceptions of

102 speech were also linked to broader psychological wellbeing (Berger and Dalton,

103 2011). The combination of self-reports and objective assessments of speech may

104 thus provide additional insight into the relationship between speech and

105 psychological adjustment.

106 Several potentially influential background or mediating factors may affect the

107 development of speech and language skills, as well as psychological adjustment,

108 including hearing problems, cognitive function, a different mother-tongue, cleft type

109 and gender (Kuehn and Moller, 2000, Millard and Richman, 2001; Flynn et al., 2009;

110 Ponduri et al., 2009; Roberts et al., 2012; Feragen et al., 2014). Cognitive function

111 may further be related to the presence of other medical and/or psychological

112 conditions additional to the cleft, such as learning difficulties, attention

113 deficit/hyperactivity disorder (AD/HD), autism spectrum disorder, dyslexia, specific

114 language impairment and developmental delay (Feragen et al., 2014). It is therefore

115 necessary to identify such underlying factors, to the extent possible, in order to

116 control their impact on the chosen outcome variables. This information should thus

117 be registered and methodologically controlled when investigating language and

118 reading skills in children with CL/P.

119 In summary, while deficits in speech development and reading ability have been

120 found to be prevalent in children with CL/P, there has been less research on

121 language development and/or how measures of language skills relate to speech and

122 reading in this patient group (Hardin-Jones and Chapman, 2011). Further,

123 information concerning the ways in which all three may impact upon psychological

124 variables is scarce. In both the general population (Goodyer, 2000; Conti-Ramsden

125 et al., 2013; Knivsberg, 2012) and in relation to children with CL/P (Berger and

126 Dalton, 2011; Richman et al., 2012), concerns have been raised regarding the

127 psychological vulnerability of those with speech, reading and language difficulties.

128 Since competency with spoken and written language is important for a child's

129 success both in school and beyond (Chapman, 2011), and given the value placed on

130 educational achievement in western societies, this paucity of information is

131 concerning. There is a need for new research, which specifically addresses the

132 relationship between speech, reading, language and psychological adjustment,

133 research that should take self-reported satisfaction with speech into account and

134 involve collaborative efforts between psychologists and speech and language

135 therapists.

136 The aim of the present study was to explore possible associations between

137 psychological variables and measures of speech, language and reading, in order to

138 explore markers of psychological risk in children with cleft. Associations were

139 investigated between validated and objective measures of speech, language,

140 reading and:

141   1) Psychological adjustment (self- and parent reports)

142   2) Subjective satisfaction with speech

143   3) Perceived teasing (self- and parent reports)

144

145 **Method**

146 **Design and participants**

147 The current study was based on multidisciplinary cross-sectional clinical data from

148 children born with two different cleft types: cleft lip and palate (CLP) or cleft palate

149 only (CP), aged ten at the time of routine speech, language, and psychological

150 assessment. Norway provides centralised treatment of cleft, and most of the children

151   were treated according to the Oslo cleft team protocol, which involves palate repair

152   at 12-14 months.

153   Three birth cohorts of children, born in 2000, 2002, and 2003, were included in the

154   study (*n* = 170). Sample attrition since birth included seven children (*n* = 7/177): four

155   due to death, two had moved out of the country, and one family did not want any

156   follow-up from the team. There were 78 children with CLP and 92 with cleft palate.

157   Further, there were 99 boys (58%) and 71 girls (42%).

158   Parents were 55% mothers (n = 93), 21% fathers (n = 36), or both parents together

159   (n = 38, 22%). Three respondents (2%) were not the child's parents, and included

160   grandparents or foster parents.

161   For the first birth cohort, language and reading tests were not performed on adopted

162   children, children that did not have Norwegian as their first language, and children

163   with diagnosed conditions in addition to the cleft (n = 33). No measures of language

164   and reading were administered on the 2001 birth cohort, and reading skills were not

165   assessed for the first 23 children of the 2003 cohort, both due to changes in

166   protocols at the time of assessments. Therefore, there is some variation in the

167   sample size regarding some variables (see Table 1). In addition, five children were

168   not able to undergo the routine evaluations of speech, language or reading, and/or to

169   complete the psychological self-reported questionnaires, due to severe

170   developmental problems.

171   The study conformed to guidelines provided by the local ethics committee (Region

172   Oslo - East). Informed consent was sought from the parents of all participants (n =

173   170). Participation rate was 100%.

Additional conditions and difficulties

Information about the presence of an additional condition or diagnosis was collected from the child's treatment records and/or from information provided by the parents at the time of assessment and/or by the local health services. Due to a centralised treatment setting, the child is seen by the same treatment team from birth until late adolescence, and information about the presence of other difficulties and/or diagnosed conditions are thought to be highly reliable. Additional diagnoses included a wide range of conditions, such as developmental difficulties (e.g. autism spectrum disorder, developmental delay or non-specific developmental difficulty affecting the child's cognitive capacities and learning), AD/HD, specific language impairment (SLI), and dyslexia. Additionally, some children had a diagnosed syndrome, such as Treacher Collins, Opitz, or 22q11.2 deletion, with or without other associated difficulties, as described above. A description of the types and numbers of patients affected by other conditions are presented in Table 1.

Hearing problems

Information about previous or current hearing difficulties was drawn from the children's case records. Children were classified according to whether they had hearing aids, and whether they still had grommets at age 10 or other hearing difficulties. Descriptives regarding hearing difficulties are found in Table 1.

Measures

Validated measures of speech, language and reading were administered by specialist speech and language therapists, while psychological measures were administered by the team's clinical psychologists.

Speech

198   The Swedish Articulation and Nasality Test (SVANTE, Lohmander et. al., 2005) is a

199   standardised test for the assessment of articulation and nasality in children with

200   structural and/or physiological deviations such as CL/P. The measure has been

201   developed according to international standards (Lohmander et al., 2005) and the

202   Norwegian version (SVANTE-N) was used in the present study. The test includes

203   assessment of words, sentences and spontaneous speech production, and is

204   designed to systematically assess articulation and nasal resonance. In the present

205   study, the speech therapist's perceptual evaluation of Resonance (0-4) and

206   Intelligibility (0-2) according to the SVANTE's guidelines was used. All speech

207   therapists had extensive experience evaluating children with velopharyngeal

208   inadequacy and cleft. Both variables were dichotomized, categorizing children as

209   having either no problems (Resonance: 0-1; Intelligibility: 0) or mild-severe problems

210   on Resonance (2-4) and Intelligibility (1-2).

211   ***Language***

212   Language 6-16 (Språk 6-16, Ottem and Frost, 2010) is a well-established and

213   standardised screening test of language skills in children aged 6 to 16. Language 6-

214   16 includes three compulsory subscales evaluating Sentence recall, Serial recall,

215   and Vocabulary, in addition to the optional subscale Phonological Awareness.

216   Sentence Recall measures the ability to organize and retain sentences, while Serial

217   Recall is a measure of phonological short-term memory. Vocabulary evaluates the

218   semantic aspects of language. The first three subscales are summarized as a Total

219   language screening score. Phonological Awareness measures the child's

220   understanding of the rule-based sound system of the language. Each item is scored

221   as correct or incorrect (0-1) and testing within a subscale is halted after three failed

222   items. Raw scores on each subscale are converted to standard scores with a mean

223   of 10 and a standard deviation of 3. The Total language screening score has a mean

224   of 100 and a standard deviation of 15. Reliability has been reported as good on all

225   subscales (α = .71-.89) and excellent on the Total language screening score (α =

226   .91). The scale has been shown to possess good content and criterion validity in

227   addition to a coherent factor structure (Ottem and Frost, 2010). The Total Score,

228   Sentence Recall, Serial recall, and Vocabulary of the Language 6-16 has been

229   shown to correlate well with two of the subscales of the WISC-III (Wechsler, 1991):

230   Verbal Comprehension and Freedom from Distractibility (Ottem, 2007).

231   ### *Reading*

232   The Word Chain Test (Ordkjedetesten*,* Høien and Tønnesen, 2007) is a well-

233   established standardised screening test which measures phonological decoding

234   skills in children from the age of 8. Raw scores are converted to Stanine scores (1-

235   9), therefore with a mean of five and a standard deviation of two. Validity was

236   deemed satisfactory, and test re-test reliability was reported to be .84 in 10-year-old

237   children, while split-half reliability was r = .99, p < .001 (Høien and Tønnesen, 2007).

238   The Reading Comprehension Test, S-40 (Setningsleseprøven*,* Høien et al., 2008) is

239   a standardised and well-established screening test of reading comprehension in

240   children aged 9 to 16. As the Word Chain Test, raw scores are converted to Stanine

241   scores. Internal reliability was shown to be α = .90 in a sample of 11-12 year old

242   children (Høien et al., 2008).

243   ### *Psychological adjustment*

244   *Psychological and emotional adjustment:* The Strengths and Difficulties

245   Questionnaire (SDQ*;* www.sdqinfo.com; Goodman, 1997) is a screening tool for

246   strengths and behavioural difficulties in children and adolescents. The SDQ was

247 completed by one or both parents and the child in the current study. The SDQ

248 includes five subscales measuring emotional distress, conduct problems,

249 hyperactivity/attention difficulties, peer relationship problems, and pro-social

250 behaviour. Each subscale consists of five items that are positively or negatively

251 worded. Each item is scored "not true", "somewhat true" or "certainly true" (0-2). The

252 first four subscales are summarized as a Total Difficulties Score (including in total 20

253 items, with scores ranging from 0-40). Internal reliability (Cronbach's alpha) was

254 satisfactory for the Total Difficulties Score for both the child and the parent version of

255 the questionnaire (α = .77 and .84) but was modest for some of the subscales:

256 emotional distress (α = .66 and .65), conduct problems (α = .48 and .58),

257 hyperactivity/attention difficulties (α = .58 and .80), peer relationship problems (α =

258 .51 and .64) and prosocial behavior (α = .65 and .62). Similar measures of reliability

259 have been reported in previous studies (Goodman, 2001; Van Roy et al., 2008).

260 *Subjective satisfaction with speech:* The Satisfaction with Appearance Scales (SWA,

261 developed by the Psychology Special Interest Group of the Craniofacial Society of

262 Great Britain and Ireland) evaluates satisfaction with cleft-related and non-cleft-

263 related parts of the face, speech, overall appearance and visibility of the cleft

264 (Cronbach's α = .88 for the scale's 15 items). Each rating is made on an interval

265 scale of 0 to 10 where a score of 10 indicates very high levels of satisfaction. One

266 item measures the child's satisfaction with speech ("*How satisfied are you with your*

267 *speech (=the sounds you make when you speak)?*"), and was used in the current

268 study.

269 *Self-reported teasing*: Subjective experiences of teasing were measured through the

270 Child Experience Questionnaire (CEQ, Pertschuk and Whitaker, 1982). The CEQ

271 utilizes a five-point Likert scale to reflect the child's self-report of positive and

272   negative social experiences, with high scores reflecting positive social experiences.

273   One item measures perceived teasing and was used in the present study ("*I am*

274   *teased*"). The child's reports of teasing (five-point Likert scale) were further

275   categorised into three groups: never/very seldom, sometimes, and often/very often.

276   *Parent-reported teasing:* Parents completed the Parent Questionnaire (developed by

277   the Psychology Special Interest Group of the Craniofacial Society of Great Britain

278   and Ireland). The questionnaire includes a question about whether the parents

279   believe the child is currently being teased or not (dichotomy; "*Has teasing or bullying*

280   *been a problem for your child?*").

281   **Statistical analysis**

282   Analyses were performed using SPSS 22 and AMOS 22 (IBM Corp, Armonk, NY).

283   Preliminary analyses were undertaken to investigate the role of the potentially

284   influential background variables gender, cleft type, hearing problems, a different

285   mother-tongue, and the presence of an additional condition on language and reading

286   scores. In order to control for an accumulation of Type I errors, as would be the case

287   with successive *t*-tests, one-way ANOVA was chosen for this purpose. Since each

288   outcome variable was tested with regards to five background variables, F-statistics

289   from the ANOVA are reported in range mode to enhance readability. When exploring

290   the impact of an additional condition on objective measures of speech, chi-square

291   tests were performed.

292   Following the analyses on background factors, a path analysis was used to test for

293   the impact of language, reading, and speech on psychological adjustment and on the

294   child's subjective satisfaction with speech. Only the background variable having

295   been shown to significantly impact on language and reading was included in the path

analysis. In order to keep statistical strength to a maximum, only the total scores (Total Language Screening scale and the Total SDQ score) were used, in addition to the other main variables. Following recommendations in the AMOS users' guide (Arbuckle, 2007) model fit was determined using several indices. Model fit criteria were χ^2 (should not be significant), the Normed Fit Index, NFI, and the Comparative Fit Index, CFI (both should be higher than 0.95, acceptable above 0,90), the Root Mean Square Error of Approximation, RMSEA (should be lower than 0.06, acceptable if lower than 0.08), and its lower (Lo90) and upper (Hi90) ends of a 90% confidence interval.

In order to investigate the associations between speech, language, reading, and psychological measures in more detail, correlations (Pearson's *r*) were calculated for the subscales of the SDQ and language, reading, and speech. Since the subscale Phonological Awareness is not included in the Total Language Screening score, this subscale was included in these more explorative analyses.

Last, and in order to check whether experiences of teasing were related to language, reading, and speech difficulties, analysis of variance with Tukey multiple-comparison tests (self-reports), and independent sample t tests (parent reports) were performed.

Results

Sample characteristics and descriptives are provided in Table 1, including information about therapist-rated assessment of problems with resonance and intelligibility, and language and reading scores for the total sample.

Preliminary analyses

318   As expected, the impact of an additional condition (such as a syndrome,

319   developmental difficulties or delay, AD/HD, SLI, and dyslexia) was highly significant

320   on all outcome measures (Reading Comprehension: $F_{(7,102)} = 25.47$, $p < .001$;

321   Word Chain: $F_{(7,102)} = 22.38$, $p < .001$; Total Language Screening: $F_{(7,118)} =$

322   26.50, $p < .001$; Sentence Recall: $F_{(7,118)} = 19.45$, $p < .001$; Serial recall: $F_{(7,119)}$

323    $= 20.82$, $p < .001$; Vocabulary: $F_{(7,118)} = 15.06$, $p < .001$; Phonological Awareness:

324    $F_{(7,111)} = 28.96$, $p < .001$).

325   Analyses also indicated an association between the presence of other conditions

326   additional to the cleft and intelligibility. While 82.3% (n = 51) of the children with no

327   additional condition had normal intelligibility scores, this was only the case in

328   approximately half of the children with an additional condition (52.8%, n = 19; $\chi^2 =$

329   9.70, p < .01). Resonance, however, was not related to the presence of conditions

330   additional to the cleft. A total of 63.5% of the children with cleft and no additional

331   condition had resonance scores within the normal range, compared to 52.9% of the

332   children with an additional condition ($\chi^2 = 1.02$, p > .05).

333   The other background factors did not impact significantly on language and reading

334   scores (Reading Comprehension: $F_{(7,102)} = 0.04 - 1.70$, $p > .05$; Word Chain: F

335    $(7,102) = 0.06 - 2.25$, $p > .05$; Total Language Screening: $F_{(7,118)} = 0.02 - 3.75$, $p >$

336   .05; Sentence Recall: $F_{(7,118)} = 0.00 - 2.47$, $p > .05$; Serial recall: $F_{(7,119)} = 0.44 -$

337   1.26, $p > .05$; Vocabulary: $F_{(7,118)} = 0.02 - 1.17$, $p > .05$; Phonological Awareness:

338    $F_{(7,111)} = 0.09 - 2.67$, $p > .05$).

339   Since none of the background factors impacted significantly on the variables, except

340   for the presence of an additional condition, only this last variable was taken into

341   account in the subsequent analyses. Associations were not expected to differ

according to whether the child had or did not have a condition additional to the cleft. In order to check for this assumption, all analyses were run separately for children with and without an additional condition in preliminary analyses. Results indicated that the associations between the variables were not consequently affected by differences in means. Subsequent analyses were therefore presented for the total sample, in order to increase statistical strength.

Associations between speech, language, reading and psychological variables

Self-reported psychological adjustment

The hypothesized model (Model 1) of potential associations between language, reading, speech and psychological variables was tested in AMOS. Goodness-of-fit statistics indicated a moderate fit (χ^2 (15, n = 170) = 27.84, p = .023; CFI = 0.92; NFI = 0.86; RMSEA = 0.071, Lo90 = 0.026, Hi90 = 0.112). The path analysis revealed that some regression weights were not statistically significant (Intelligibility and subjective satisfaction with speech; Total language screening score and the SDQ Total score). Therefore, in the corrected model (Model 2), these two associations were deleted. Goodness-of-fit statistics indicated a better fit (χ^2 (17, n = 170) = 28.94, p = .035; CFI = 0.93; NFI = 0.86; RMSEA = 0.064, Lo90 = 0.017, Hi90 = 0.104). The path estimates and explained variances are provided in Figure 1. As also demonstrated through the preliminary analyses, all path estimates between the presence of an additional condition and measures of speech, language and reading were highly significant (p < .001). In addition, and as expected, there were clear associations between language scores and assessments of reading (p < .001). However, while language scores did not directly predict self-reports of psychological adjustment, reading skills did (p < .05). Further, problems with resonance were positively correlated with intelligibility (p < .001), and negatively with the child's self-

reported satisfaction with speech (p < .05). However, only 7% of the variance in subjective satisfaction with speech was explained by objective measures of speech (R^2 = .07). Lastly, subjective satisfaction with speech was associated with psychological adjustment (p < .01). In summary, 20% of the variance in psychological adjustment was directly explained by reading skills and the child's subjective satisfaction with speech, and indirectly by language development and objective measures of speech (R^2 = .20).

Parent-reported psychological adjustment

The same hypothesized model as for self-reports was tested in AMOS, indicating less satisfactory goodness-of-fit statistics than for self-reports (χ^2 (13, n = 170) = 34.35, p = .001; CFI = 0.88; NFI = 0.84; RMSEA = 0.099, Lo90 = 0.059, Hi90 = 0.139). The path analysis revealed several non-significant regression weights in the original model. No significant associations were found between Resonance, Language Screening, Intelligibility, Subjective satisfaction with speech, and the SDQ. In addition, the link between Intelligibility and Subjective satisfaction with speech was also non-significant. Therefore, in the corrected model (Model 2), these associations were deleted. Goodness-of-fit statistics were recalculated, and indicated a slightly better fit, however still moderate (χ^2 (18, n = 170) = 34.44, p = .003; CFI = 0.88; NFI = 0.82; RMSEA = 0.082, Lo90 = 0.046, Hi90 = 0.118). Since language, reading and objective speech assessments were the same as in the model that tested self-reported adjustment, path estimates and explained variances for these variables are the same as those in Figure 1. The main difference between the model based on self-reports compared to parent reports was that the child's subjective satisfaction with speech did not predict parent-reported psychological adjustment (p > .05). A minor difference was also found in the strength of associations between reading and

392   parent reported psychological adjustment (Reading Comprehension = -.22; Word

393   Chain test = -.26; p < .05). In summary, 15% of the variance in parent-reported

394   psychological adjustment was directly explained by reading skills, and indirectly by

395   language development. There were no significant associations between objective

396   measures of speech and psychological adjustment according to parent reports (p >

397   .05).

398   ***Subscales of the SDQ: Associations with language, reading and speech***

399   In order to further investigate the impact of language, reading, and speech difficulties

400   on psychological adjustment, correlations including the SDQ's subscales (emotional,

401   cognitive, behavioural and social adjustment) were calculated. Results for self-

402   reports are given in Table 2, while the results for the parent reports are found in

403   Table 3. The Total Score of the SDQ was significantly associated with language and

404   reading subscales for self-reports (*r* = -.20 to -.27) and parent reports (*r* = -.19 to -

405   .33). A similar pattern was evident for self-reported Emotional Distress (*r* = -.23 to -

406   .25), and parent-reported Social Difficulties (*r* = -.23 to -.27). Interestingly, measures

407   of language and reading correlated with neither parent-reported Emotional Distress

408   nor self-reported Social Difficulties. The Language Screening Total score was also

409   associated with problems of attention and/or hyperactivity based on both self-reports

410   (*r* = -.22 to -.24) and parent reports (*r* = -.22 to -.36).

411   As can be seen from Tables 2 and 3, the associations between the objective speech

412   variables and the SDQ were non-significant for all subscales, except for the

413   associations between Intelligibility and parent-reported Total Problem Scores (*r* =

414   .22, *p* < .05) and Social Difficulties (*r* = .31, *p* < .01). Subjective satisfaction with

415   speech on the other hand, correlated significantly in self-reports for all subscales on

416   the SDQ except Social Difficulties. There were no significant associations between

417   the parent-reported SDQ and the child's subjective evaluations of speech.

418   ***Teasing: self- and parent reports***

419   Most children reported no or very few experiences of teasing (79.1%, n = 76), while

420   17.7% (n = 17) described it to happen sometimes. Only 3.1% (n = 3) of the children

421   said they were teased often or very often. According to parent reports, 65.8% (n =

422   73) were not teased, while 34.2% had experienced teasing (n = 38). Self- and parent

423   reports of teasing correlated relatively well (*r* = .51, p < .01, *n* = 109).

424   As can be seen in Table 4, the more reported teasing, the lower language and

425   reading skills, except for Reading Comprehension. Children reporting frequent and

426   repeated teasing had language and reading scores (Word Chain Test) within the

427   lower the normal range or below, while children who said they were never or seldom

428   teased had language and reading scores within the normal range. However, Tukey

429   Post Hoc analyses revealed that none of these differences were statistically

430   significant.

431   Associations between parent-reported teasing and language and reading skills are

432   provided in Table 5 and reveal that children who were teased had lower scores on

433   both reading tests. However, differences were only statistically significant for the

434   Word Chain test (*t* (92) = 2.05, p < .05). There were no differences in language

435   scores between the two groups.

436   There seemed to be fewer problems with intelligibility in children who reported little

437   or no teasing (Table 4). However, this difference was not statistically significant, and

438   the mean score for those few children reporting repeated teasing indicated only mild

439   problems with intelligibility in this group. There were no associations between parent

440   reports of teasing and intelligibility. Calculations of means regarding resonance was

441   neither associated with self-reported (Table 4) nor parent-reported teasing (Table 5).

442   Subjective satisfaction with speech was significantly associated with parent-reported

443   teasing (t (86) = 2.53, p < .05), while the differences between the groups in self-

444   reported teasing were not statistically significant. However, children reporting teasing

445   'sometimes' were less satisfied with their speech than children who did not

446   experience any or almost any teasing. Unexpectedly, the few children (n = 3)

447   reporting repeated teasing were very satisfied with their speech.

448

449   **Discussion**

450   This study explored associations between speech, language, reading and

451   psychological adjustment, including measures of teasing and subjective satisfaction

452   with speech. Self-reports, parent-reports and assessments carried out by specialist

453   speech and language therapists and clinical psychologists were included. Analyses

454   indicated associations between reading skills and psychological adjustment, as well

455   as associations between subjective satisfaction with speech and psychological

456   adjustment. Further, results indicated a possible association between experiences

457   of teasing and some measures of language, reading and speech. Differences

458   between 'objective' assessments and subjective reports were observed.

459   **Language, reading, and psychological adjustment**

460   While problems with language were not directly related to psychological adjustment

461   in the path analysis, difficulties with reading (both self- and parent report) were. This

462   finding may point to the importance placed on reading skills in western societies.

463 According to the correlational analysis, language and reading were associated with

464 emotional difficulties (self-report), social problems (parent-report) and difficulties with

465 attention/hyperactivity (self- and parent reports).

466 In a society where literacy is a highly valued skill, children's feelings of competence

467 and emotional well-being may be shaped by the comparisons they make between

468 themselves and others (Burden, 2008). As the present findings suggest, difficulties in

469 language and reading may subsequently contribute to emotional distress. Self-

470 reported emotional difficulties have also been linked to language and reading skills in

471 the general population (Arnold et al., 2005; Terras et al., 2009) and although little is

472 known about the underlying factors, several hypotheses have been offered

473 (Maughan and Carroll, 2006). For example, co-morbidity may be explained by

474 common risk factors, such as neurobiological factors, but also by a causal link,

475 whereby reading difficulties may increase the likelihood of emotional problems.

476 Another hypothesis proposes attentional deficits as a potential underlying factor

477 (Carroll et al., 2005). Although the cross-sectional design of the present study

478 prevents us from drawing conclusions about causality, the findings confirm that

479 psychological variables, such as emotional adjustment and attention, are associated

480 with language and reading skills, which could be related to underlying

481 neurobiological components (Richman and Ryan, 2003; Nopoulos et al., 2010;

482 Conrad et al., 2014).

483 **Speech problems and psychological adjustment**

484 The path analysis indicated that participants' subjective satisfaction with speech was

485 not associated with the speech therapists' assessments of intelligibility, or with

486 objective measures of language and reading. This is in line with Conrad et al. (2014)

487 who did not find associations between reading and measures of speech, and with

488 Havstam et al. (2008), who only found weak associations between subjective and

489 'objective' measures of speech in adults with a cleft. However, a significant

490 association was found between the speech therapist's assessment of resonance and

491 the child's satisfaction with speech, suggesting that children with CL/P may be aware

492 of potential hypernasality in their speech at age 10.

493 The only 'objective' speech variable that correlated with psychological measures

494 according to correlational analyses was intelligibility, which was associated with

495 parent-reported general psychological difficulties and social problems. This

496 association could suggest a psychological vulnerability in cases of certain cleft-

497 related speech problems. However, this association was not confirmed in the path

498 analysis, which may be due to other variables not accounted for in the correlational

499 analyses. This finding could, for example, be related to and/or partly explained by the

500 relationship between intelligibility and the presence of an additional condition, where

501 a higher frequency of children with an additional condition had problems related to

502 intelligibility. This relationship was supported by the path analysis and has also been

503 reported in a previous study (Persson et al., 2002). It could be that the presence of

504 an additional condition moderates the associations between intelligibility and social

505 risk. Future research is needed in order to further examine the associations between

506 an additional condition, intelligibility, and psychological risk.

507 **Teasing: self-reports and parent reports**

508 While mean scores indicated an association between the child's experience of being

509 teased and lower scores on measures of language, reading and speech, few of

510 these associations were found to be significant. This may be due in part to the

511 relatively small number of children reporting repeated experiences of being teased.

512   One of the measures of reading skills did vary significantly with parent-reported

513   teasing, an association that has been reported in a non-cleft sample (Terras et al.,

514   2009).   However, according to the present study's parent reports, mean reading

515   scores for children who were teased were still within the normal range, indicating that

516   parent reported teasing was probably not related to poor reading skills. Self-reported

517   teasing, on the other hand, was associated with language and reading difficulties,

518   possibly reflecting the child's awareness of problems with communication and their

519   potential consequences on social interaction and experiences.

520   In relation to speech, 'objective' measures of intelligibility and resonance were not

521   significantly associated with reports of negative social experiences, in line with a

522   previous study on children with CL/P (Murray et al., 2010).   In contrast, a recent

523   study found an association between problems of resonance and expected negative

524   social judgements (Watterson et al., 2013), which could further be indicative of

525   teasing experiences.   Similarly, several previous studies have reported a strong

526   association between subjectively measured speech problems and self-reported

527   teasing (Turner et al., 1997; Hunt et al., 2006), while other studies have investigated

528   patient's belief about the source of teasing, pointing to speech difficulties as a

529   potential vulnerability factor (Semb et al., 2005; Noor and Musa, 2007; Havstam et

530   al., 2011).

531   The present results illustrate the complex relationship between subjective and

532   objective outcome measures. Perceptions of teasing may be coloured by the child's

533   psychological vulnerability or strength (Snyder and Pope, 2003). Questions by peers

534   or strangers about the cleft may be experienced as teasing by a vulnerable child,

535   while a more secure child will interpret it as positive curiosity or as a simple question

536 (Feragen et al., 2009; Shavel-Jessop and Shearer, 2013). In addition, children who

537 are aware of poor language or reading skills may feel socially vulnerable, a

538 susceptibility that could be potentially strengthened if the child feels uncomfortable

539 about a visible and/or audible difference due to CL/P. In addition, the results highlight

540 the importance of independent observers, since the children's subjective

541 experiences may differ from those reported by their parents (Turner et al., 1997), as

542 was shown in the present study. More research is needed in order to further explore

543 the relationships between language and reading skills, speech quality, and social

544 vulnerability, recognised by both the parents and the child in the present study.

545 **The influence of background variables**

546 Several children participating in this study had one or more conditions in addition to

547 the cleft, such as learning difficulties, attention deficit/hyperactivity disorder (AD/HD),

548 autism spectrum disorder, dyslexia, specific language impairment and

549 developmental delay. Preliminary and path analyses demonstrated the high

550 prevalence and significant influence of an additional condition on measures of

551 language, reading and intelligibility. While the present study did not primarily aim to

552 investigate the impact of additional conditions, previous research has reported a

553 relationship between these types of conditions and speech, psychological and

554 academic outcomes (Persson et al., 2002; Feragen et al., 2014; Knight et al., in

555 press).

556 Consequently, the role of additional conditions should be considered when

557 examining the results from the present study. The findings point to a potential

558 double-association between psychological vulnerability and problems of language,

559 reading and intelligibility in this subgroup of children with CL/P, as has been

560 documented in the non-cleft population (Bishop, 2009). Since children with

561   conditions in addition to the cleft have been shown to be a potentially vulnerable

562   subgroup in other cleft samples (Persson et al., 2002; Feragen and Stock, 2014;

563   Knight et al., in press), the possibility of this factor being of central importance also

564   for language and reading skills in children with CL/P should be considered and

565   discussed in future studies. In addition, the impact of an additional condition on

566   speech variables requires further investigation. Recent neuropsychological research

567   has investigated associations between brain structure and behavioural outcomes in

568   young people with cleft (Conrad et al., 2010; Nopoulos et al., 2010). More

569   specifically, Conrad et al. (2014) reported an association between cerebellum size

570   and problems with articulation in boys. The authors ask whether underlying variables

571   that were not measured could explain this relationship. The findings of the present

572   study suggest that the presence of an additional condition could potentially be a

573   confounding variable in the development of speech, language, and reading. A better

574   understanding of this potentially critical background variable would help us

575   distinguish which outcomes are related to the cleft and which are associated with

576   having an additional condition(s). The findings of the current study also emphasise

577   the need to identify additional difficulties as early as possible to facilitate the initiation

578   of appropriate interventions.

579   The impact of other potentially influential background variables such as gender, cleft

580   type, different mother-tongue, hearing difficulties and secondary surgery were not

581   found to significantly affect results. However, these variables may require further

582   investigation, since associations with speech and language have been identified

583   within the cleft population (for a review, see Kuehn and Moller, 2000). In the present

584   study, these variables represented small subsamples, and thus individual variations

585   may explain the non-significant findings.

Clinical implications

The associations between language and reading, and their potential impact on emotional and social development, suggest that speech therapists and psychologists, in addition to other health professionals, teachers and parents, should be particularly alert to potential psychological difficulties in children with CL/P who have language and reading difficulties. This suggestion also applies in some respect to problems related to 'objective' ratings of intelligibility. In addition, the child's subjective satisfaction with speech may be a useful indicator of psychological risk, at least in relation to self-reported psychological adjustment.

The differences observed between self- and parent reports, as well as between 'objective' and subjective measures are interesting. Discrepancies between self- and parent reports have been described previously when using the SDQ (Van Roy et al., 2010). Such findings may be due to parents having a greater capacity than children to observe and identify social problems, while emotional difficulties may not be apparent to anyone other than the affected person. In addition, 'objective' measures of speech were not significantly associated with psychological adjustment, while participants' subjective ratings of speech were. These differences highlight the importance of including multiple perspectives during clinical assessments, in order to capture the complexity of perceptions of psychological adjustment.

The results of the present study illustrate the importance of a multidisciplinary approach to the treatment of children with CL/P, including the monitoring of speech, language and reading skills and the assessment of psychological adjustment. In addition, and given the variation in levels of care provided within some cleft teams across and within countries (Fox and Stone, 2013; Scott et al., in press 2014), the identification of variables other than those pertaining to psychological adjustment

611   that could identify children with cleft as being at risk are valuable and would allow a

612   more targeted allocation of limited resources.

613   **Strengths and limitations**

614   One of the strengths of the present study was that information was drawn from three

615   almost-complete birth cohorts, with a participation rate of 100%. Due to centralised

616   treatment, the sample can be expected to be representative of the population under

617   study. Furthermore, the sample was able to shed light on the potentially vulnerable

618   subsample of children with associated conditions, raising awareness about those

619   potentially at risk for speech, language, and reading problems, in addition to potential

620   psychological risk. Furthermore, the restricted age range reduced the possible

621   confounding variable of developmental stage. Another strength was the use of

622   validated instruments regarding language, reading and psychological adjustment. In

623   addition, speech was assessed both subjectively and rated by trained speech and

624   language therapists, providing a double-perspective on potential speech problems.

625   Additionally, psychological outcome measures were completed by both the children

626   and the parents, also strengthening the findings. The multidisciplinary approach,

627   linking validated measures of speech and language skills with psychological

628   adjustment, also add value to the present study.

629   Nevertheless, several limitations also have to be considered. First, without a control

630   group allowing for comparisons with the general population, it is difficult to tease

631   apart which findings may apply specifically to children with CL/P. Nonetheless, few

632   studies have addressed the potential impact of speech, language and reading on

633   psychological variables, and thus the present study offers an important step on the

634   way to improved knowledge. Second, while information about hearing was provided

635   and controlled for, the cross-sectional nature of the study meant that information was

636 missing in some cases. More detailed and specific information about hearing

637 difficulties are warranted. Further, future studies should also aim to include other

638 cleft-related disciplines, such as surgeons and orthodontists, in order to provide a

639 holistic perspective. A third limitation was related to the use of national measures of

640 language and reading, restricting comparisons between studies carried out in other

641 countries. The psychological outcome measure, however, is broadly used

642 internationally. Fourth, only two measures of objective speech evaluations were

643 included, and speech was assessed by the child's speech therapist only, impeding

644 calculations of internal reliability. Another measurement issue was that information

645 about teasing was provided by one item only in both self-reports and parent reports.

646 However, issues related to teasing and negative social experiences are discussed in

647 depth with the child during the psychological routine assessment when needed, and

648 are therefore believed to reflect the child's perception of his or her social

649 experiences. Finally, language assessments did not include tests of reading ability in

650 the first birth cohort, due to different team routines at the time. Nonetheless, all

651 measures were available for two complete birth cohorts. A final limitation was the

652 lack of demographic information such as socio-economic status. However, the

653 potential impact of such demographic information on the results was considered to

654 be low, given that SES and educational level are expected to have a reduced impact

655 in Norwegian samples than in many other Western societies (Heiervang et al., 2008).

656 Nevertheless, future research should aim to include such information.

657 **Conclusions**

658 Associations were identified between language, reading, speech and psychological

659 adjustment. The findings confirm the need to include both self- and parent reported

660 measures, in order to capture multiple perspectives in research and clinical

661 assessments. Cleft teams, in addition to teachers and local health services, should

662 be aware of co-variations between problems with speech, language, reading and

663 psychological difficulties, in order to identify potentially vulnerable children and

664 maximise the likelihood of appropriate treatment and interventions. Future and

665 longitudinal studies should examine cross-discipline associations further, in order to

666 gain a better understanding of which interventions may be the most suitable.

667
668
669
670 **References**

671 Arbuckle, J. L. *Amos 16.0 user's guide: SPSS*. Chicago, IL, USA: Amos

672   Development Corporation; 2007.

673 Arnold EM, Goldston DB, Walsh AK, Reboussin BA, Daniel SS, Hickman E, Wood

674   FB. Severity of emotional and behavioural problems among poor and typical

675   readers. *J Abnorm Child Psychol.* 2005;22:205-217.

676 Berger ZE, Dalton LJ. Coping with a cleft II: Factors associated with psychosocial

677   adjustment of adolescents with a cleft lip and palate and their parents. *Cleft*

678   *Palate Craniofac J.* 2011;48:82–90.

679 Bishop DVM. Genes, Cognition, and Communication. Insights from

680   neurodevelopmental disorders. The year in cognitive neuroscience. *Annals*

681   *of the New York Academy of Sciences.* 2009;1156:1–18.

682 Boes A, Murko V, Wood J, Langbehn D, Canady J, Richman L, Nopoulos P. Social

683   function in boys with cleft lip and palate: Relationship to ventral frontal cortex

684   morphology. *Behav Brain Res.* 2007;181:224–231.

685 Burden R. Is dyslexia necessarily associated with negative feelings of self-worth? A

686   review and implications for future research. *Dyslexia.* 2008;14:188-196.

687 Carroll JM, Maughan B, Goodman R, Meltzer H. Literacy difficulties and psychiatric

688   disorders: evidence for comorbidity. *J Child Psychol Psychiatry.* 2005;46:524-

689   532.

690 Chapman K. The relationship between early reading skills and speech and language

691   performance in young children with CLP. *Cleft Palate Craniofac J*. 2011;48:

692   301-311.

693 Conrad A, Dailey S, Richman L, Canady J, Karnell M, Axelson E, Nopoulos P.

694   Cerebellum structure differences and relationship to speech in boys and girls

695   with nonsyndromic cleft of the lip and/or palate. *Cleft Palate Craniofac J*.

696   2010;47:469–475.

697 Conrad AL, McCoy TE, Devolder I, Richman LC, Nopoulos P. Reading in subjects

698   with an oral cleft: Speech, hearing and neuropsychological skills.

699   *Neuropsychology.* 2014;28:415-422.

700 Conti-Ramsden G, Mok PL, Pickles A, Durkin K. Adolescents with a history of

701   specific language impairment (SLI): Strengths and difficulties in social,

702   emotional and behavioral functioning. *Res Dev Disability*. 2013;34:4161-4169.

703 Feragen KJB, Borge AIH, Rumsey N. Social experience in 10-year-old children born

704   with a cleft: exploring psychosocial resilience. *Cleft Palate Craniofac J.*

705   2009;46:65–74.

706 Feragen KJB, Stock NM. When there is more than a cleft: psychological adjustment

707   when a cleft is associated with an additional condition. *Cleft Palate Craniofac*

708   *J.* 2014;51:5-14.

709 Feragen KJB, Stock NM, Rumsey N. Towards a reconsideration of inclusion and

710   exclusion criteria in cleft lip and palate: implications for psychological

711   research. *Cleft Palate Craniofac J*. 2014;51:570-579.

712 Flynn T, Möller C, Jönsson R, Lohmander A. The high prevalence of otitis media

713   with effusion in children with cleft lip and palate as compared to children

714   without clefts. *Int J Pediatr Otorhinolaryngol*. 2009;73:1441-1446.

715 Fox LM, Stone PA. Examining the team process: Developing and sustaining effective

716   craniofacial team care. In: Berkowitz S, ed., *Cleft Lip and Palate: Diagnosis and*

717   *Management (third edition)*. Springer-Verlag: Berlin Heidelberg; 2013:885-906.

718 Goodman R. The Strengths and Difficulties Questionnaire: A research note. *J Child*

719   *Psychol Psychiatry*. 1997;38:581-586.

720 Goodman R. Psychometric properties of the Strengths and Difficulties Questionnaire,

721   *J Am Acad Child Adolesc Psychiatry*. 2001;40:1337-1345.

722 Goodyer IM. Language difficulties and psychopathology. In: Bishop DVM and

723   Leonard LB, eds., *Speech and language impairments in children: Causes,*

724   *characteristics, intervention and outcome*. Philadelphia: Psychological Press;

725   2000:227-244.

726 Hardin-Jones M, Chapman KL. Cognitive and language issues associated with cleft

727   lip and palate. *Semin Speech Lang.* 2011;32:127-140.

728 Havstam C, Laakso K, Lohmander A, Ringsberg KC. Taking charge of

729   communication: Adults' descriptions of growing up with a cleft-related speech

730   impairment. *Cleft Palate Craniofac J*. 2011;48:717-726.

731 Havstam C, Lohmander A, Dahlgren Sandberg A, Elander A. Speech and

732   satisfaction with outcome of treatment in young adults with unilateral or

733   bilateral complete clefts. *Scand J Plast Reconst Surg Hand Surg*. 2008;

734   42:182-189.

735 Heiervang E, Goodman A, Goodman R. The Nordic advantage in child mental

736   health: Separating health differences from reporting style in a crosscultural

737   comparison of psychopathology. *J Child Psychol Psychiatry.* 2008;49:678–

738   685.

739   Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and

740   palate: a systematic review. *Eur J Orthod.* 2005;27:274–285.

741   Hunt O, Burden D, Hepper P, Stevenson M, Johnston C. Self-reports of psychosocial

742   functioning among children and young adults with cleft lip and palate. *Cleft*

743   *Palate Craniofac J.* 2006;43:598-605.

744   Høien T, Tønnesen G. Ordkjedetesten [Word chain test]. Bryne, Norway:

745   *Logometrica* AS; 2007.

746   Høien T, Tønnesen G, Igland G. Setningsleseprøven [Reading comprehension].

747   Bryne, Norway: *Logometrica* AS; 2008.

748   Kapp-Simon KA, Simon DJ, Kristovich S. Self-perception, social skills, adjustment,

749   and inhibition in young adolescents with craniofacial anomalies. *Cleft Palate*

750   *Craniofac J.* 1992;29:353–356.

751   Knight J, Cassell CH, Meyer RE, Strauss RP. Academic outcomes of children with

752   isolated orofacial clefts compared with children without a major birth defect.

753   *Cleft Palate Craniofac J.* In press 2014.

754   Knivsberg, A.-M. Behaviour and attention in dyslexia. In: Alexander-Passe N, ed.,

755   *Dyslexia and mental health. Investigations from differing perspectives.*

756   London, UK: Neil Alexander-Passe; 2012:27-38.

757   Kuehn DP, Moller KT. Speech and language issues in the cleft population: The state

758   of the art. *Cleft Palate Craniofac J.* 2000;37:348-1-35.

759   Lohmander A, Borell E, Henningsson G, Havstam C, Lundeborg I, Persson C.

760   SVANTE – *Svenskt Artikulations- och Nasalitets Test* (Swedish Articulation

761   and Nasality Test) [In Swedish]. Skivarp, Sweden: Pedagogisk Design; 2005.

762 Maughan B, Carroll J. Litteracy and mental disorders. *Curr Opin Psychiatry.*

763   2006;19:350-354.

764 McWilliams BJ, Musgrave RH. Psychological implications of articulation disorders in

765   cleft palate children. *Cleft Palate J.* 1972;9:294–303.

766 Millard T, Richman LC. Different cleft conditions, facial appearance, and speech:

767   relationship to psychological variables. *Cleft Palate Craniofac J.* 2001;*38*:68-

768   75.

769 Murray L, Arteche A, Bingley C, Hentges F, Bishop DV, Dalton L, Goodacre T, Hill J,

770   Cleft Lip and Palate Study team. The effect of cleft lip on socio-emotional

771   functioning in school-aged children. *J Child Psychol Psychiatry.* 2010;51:94-

772   103.

773 Noor SNFM, Musa S. Assessment of patients' level of satisfaction with cleft

774   treatment using the Cleft Evaluation Profile. *Cleft Palate Craniofac J.*

775   2007;44:292-303.

776 Nopoulos P, Boes AD, Jabines A, Conrad AL, Canady J, Richman L, Dawson JD.

777   Hyperactivity, impulsivity, and inattention in boys with cleft lip and palate:

778   relationship to ventromedial prefrontal cortex morphology. *J Neurodev Disord.*

779   2010;2:235-242.

780 Ottem E. Språk 6-16: Nye normer til bruk I forskning og utredning [*Language 6-16:*

781   *New norms for research and assessments*]*. Skolepsykologi.* 2007;4:13-23.

782 Ottem E Frost J. Språk 6-16 Screeningtest [*Language 6-16 Screening test*]. Manual

783   III. Oslo, Norway: Bredtvet kompetansesenter; 2010.

784 Persson C, Elander A, Lohmander-Agerskov A, Søderpalm E. Speech outcomes in

785   isolated cleft palate: Impact of cleft extent and additional malformations. *Cleft*

786   *Palate Craniofac J.* 2002;39:397-408.

787 Pertschuk MJ, Whitaker LA. Social and psychological effects of craniofacial

788   deformity and surgical reconstruction. *Clin Plast Surg.* 1982;9:297–306.

789 Ponduri S, Bradley R, Ellis PE, Brookes ST, Sandy J-R, Ness AR. The

790   management of otitis media with early routine insertion of grommets in

791   children with cleft palate - A systematic review. *Cleft Palate Craniofac J.*

792   2009;46:30-38.

793 Richman LC. Behavior and achievement of cleft palate children. *Cleft Palate J.*

794   1976;13:4–10.

795 Richman LC. Self-reported social, speech, and facial concerns and personality

796   adjustment of adolescents with cleft lip and palate. *Cleft Palate J.* 1983;20:108–

797   112.

798 Richman LC, McCoy TE, Conrad AL, Nopoulos PC. Neuropsychological, behavioural

799   and academic sequelae of cleft: early developmental, school age and

800   adolescent/young adult outcomes. *Cleft Palate Craniofac J.* 2012;49:387-396.

801 Richman LC, Ryan SM. Do the reading disabilities of children with cleft fit into current

802   models of developmental dyslexia? *Cleft Palate Craniofac J.* 2003;40:154-

803   157.

804 Roberts RM, Mathias JL, Wheaton P. Cognitive functioning in children and adults

805   with nonsyndromal cleft lip and/or palate: A meta-analysis. *J Pediatr Psychol.*

806   2012;37:786-797.

807 Rumsey N, Stock NM. Living with a cleft: psychological challenges, support and

808   intervention. In: Berkowitz S, ed., *Cleft Lip and Palate: Diagnosis and*

809   *Management.* 3rd ed. Berlin, Germany: Springer-Verlag; 2013:907-915.

810 Sandy J, Rumsey N, Persson M, Waylen A, Kilpatrick N, Ireland T, Ness A. Using

811   service rationalisation to build a research network: Lessons from the

812   centralisation of UK services for children with cleft lip and palate. *Br Dent J.*

813   2012;212:553-555.

814   Sandy J, Williams A, Mildinhall S, Murphy T, Bearn D, Shaw B, Sell D, Devlin B,

815   Murray J. The Clinical Standards Advisory Group (CSAG) cleft lip and palate

816   study. *British J Ortho.* 1998;25:21-30.

817   Scherer, N.J., Williams, A.L., Proctor-Williams, K. Early and later vocalization in

818   children with and without cleft palate. *Int J Pediatr Otorhinol.* 2008;72:827-

819   840.

820   Scott JK, Leary SD, Ness AR, Sandy JR, Persson M, Kilpatrick N, Waylen AE.

821   Centralization of services for children born with orofacial clefts in the

822   United Kingdom: A cross-sectional survey. *Cleft Palate Craniofac J.* In press.

823   Semb G, Brattström V, Mølsted K, Prahl-Andersen B, Zuurbier P, Rumsey N, Shaw

824   WC. The Eurocleft study: Intercenter study of treatment outcome in patients

825   with complete cleft lip and palate. Part 4: Relationship among treatment

826   outcome, patient/parent satisfaction, and the burden of care. *Cleft Palate*

827   *Craniofac J.* 2005;42:83-92.

828   Shavel-Jessop S, Shearer J. Managing teasing and bullying. In: Berkowitz S, ed.,

829   *Cleft Lip and Palate: Diagnosis and Management.* 3rd ed. Berlin, Germany:

830   Springer-Verlag; 2013:917-926.

831   Simonds JF, Heimburger RE. Psychiatric evaluation of youth with cleft lip palate.

832   *Cleft Palate J.* 1978;15:193–201.

833   Snyder HT, Pope AW. Presenting a different face to the world: Social comparisons

834   and self-evaluation of children with a facial difference. *The Psychotherapy*

835   *Patient.* 2003;12:155–174.

836 Terras MM, Thompson LC, Minnis H. Dyslexia and psychosocial functioning: An

837   exploratory study of the role of self-esteem and understanding. *Dyslexia.*

838   2009;15:304-327.

839 Turner SR, Thomas PWN, Dowell T, Rumsey N, Sandy JR. Psychological outcomes

840   amongst cleft patients and their families. *Br J Plast Surg.* 1997;50:1-9.

841 Van Roy B, Grøholt B, Heyerdahl S, Clench-Aas J. Understanding discrepancies in

842   parent-child reporting of emotional and behavioural problems: Effects of

843   relational and socio-demographic factors. *BMC Psychiatry.* 2010;10:56.

844 Van Roy B, Veenstra M, Clench-Aas J. Construct validity of the five-factor Strengths

845   and Difficulties Questionnaire (SDQ) in pre-, early, and late adolescence. *J*

846   *Child Psychol Psychiatry.* 2008;49:1304-1312.

847 Watterson T, Mancini MC, Brancamp TU, Lewis KE. Relationship between the

848   perception of hypernasality and social judgments in school-aged children.

849   *Cleft Palate Craniofac J.* 2013;50:498-502.

850 Wechsler, D. Wechsler Intelligence Scale for Children – III. San Antonio:

851   Psychological Corp; 1991.

852

853
854
855
856 Figure 1. Standardised path estimates for Model 2 exploring associations between
857 validated measures of language, reading, speech and two psychological outcome
858 variables: self-reported satisfaction with speech and self-reported psychological
859 adjustment on the Strength and Difficulties Questionnaire (SDQ, Total score).
860
861
862
863
864
865 Note to the figure: Additional condition (No=0; Yes=1); Language and reading: Lower
866 scores indicate more problems; Resonance and Intelligibility: Higher scores indicate
867 more problems; SDQ: Higher scores indicate more problems; Subjective satisfaction
868 with speech: Lower scores indicate less subjective satisfaction.

Table 1. Study sample (*n* = 170) with demographic and background variables, in addition to means for measures of speech, language and reading.

Cleft type	n	%
Cleft lip and palate	78/170	45.9
Cleft palate	92/170	54.1
Gender		
Boys	99/170	58.2
Girls	71/170	41.8
Additional conditions[1]	60/170	35.3
Syndrome	24/170	14.1
Developmental difficulties	24/166	14.5
AD/HD	19/170	11.7
SLI and/or dyslexia	15/170	9.0
Adopted children	13/170	7.8
Different first mother-tongue	21/170	12.4
Hearing problems[2]	26/157	16.6
Hearing aids	7/157	4.5
Grommets < age 10	98/129	76.0
Grommets at age 10	5/116	4.3
Secondary surgery		
Surgery before age 10	47/139	33.8
Waiting list for surgery	4/170	2.4
Resonance		
No difficulties	58/97	59.8
Mild problems	31/97	32
Moderate/severe	8/97	8.2
Intelligibility		
No difficulties	70/98	71.4
Mild problems	24/98	24.5
Moderate/severe	4/98	4
Reading		*M (SD)*
Reading Comprehension	109/114	4.8 (1.86)
Word Chain Test	109/114	5.8 (1.92)
Language		
Total score	129/137	93.4 (15.91)
Phonological awareness	122/137	9.0 (3.12)

Note: Due to some missing data, information about sample size is specified for each variable.

---

[1] The number of children with a specific condition does not add up to the total number of children with an additional condition since a) some children had more than one additional condition, and b) some children with a diagnosed syndrome did not have any other associated difficulties.

[2] Children with hearing aids and grommets at age 10 were included in the group called Hearing problems.

Table 2. Subscales of the Strengths and Difficulties Questionnaire (self-reports): Correlations with measures of reading, language and speech.

	Total score	Emotional	Conduct	Attention	Social
Reading					
Reading Comprehension	-.28**	-.23*	-.18	-.22*	-.10
Word Chain	-.27**	-.19	-.16	-.24*	-.13
Language					
Total Language Screening	-.20*	-.25**	-.09	-.02	-.17
Phonological Awareness	-.27**	-.13	-.26**	-.24*	-.13
Speech					
Resonance	.05	.00	.02	.01	.13
Intelligibility	.14	.09	.01	.08	.21
Subjective speech (self-report)	-.29**	-.20*	-.23*	-.21*	-.15

Note: *p < .05, **p < .01.

Table 3. Subscales of the Strengths and Difficulties Questionnaire (parent reports): Correlations with measures of reading, language and speech.

	Total score	Emotional	Conduct	Attention	Social
Reading					
Reading Comprehension	-.32**	-.16	-.21*	-.36***	-.16
Word Chain	-.33**	-.13	-.21*	-.33**	-.23**
Language					
Total Language Screening	-.23**	-.11	-.21*	-.17	-.27**
Phonological Awareness	-.19*	-.03	-.10	-.22*	-.19
Speech					
Resonance	.17	.05	.14	.19	.12
Intelligibility	.22*	.12	.02	.17	.31**
Subjective speech (self-reports)	-.10	-.07	-.10	-.09	-.04

Note: *$p < .05$, **$p < .01$, *** $p < .001$.

Table 4. Self-reported experiences of teasing, with corresponding means (M) and standard deviations (SD), on validated measures of language, reading, and speech, in addition to subjective satisfaction with speech.

	Never/seldom	Sometimes	Often/very often
	% (n)	**% (n)**	**% (n)**
Reported teasing (self-reports)	79.1 (76)	17.7 (17)	3.1 (3)
Language	**M (SD)**	**M (SD)**	**M (SD)**
Total Language score	95.1 (14.21)	87.1 (21.08)	78.0 (25.71)
Reading			
Reading Comprehension	5.0 (1.75)	4.5 (2.24)	6.0 (0.00)
Word Chain Test	6.1 (1.79)	5.7 (1.80)	4.0 (1.41)
Speech			
Resonance	.47 (.65)	.43 (.51)	1.0 (0.00)
Intelligibility	.24 (.47)	.46 (.52)	1.0 (0.00)
Subjective speech (self-reports)	8.3 (2.13)	7.9 (2.92)	10.0 (0.00)

Note: Tukey Post hoc analyses between groups were all non-significant.

Table 5. Parent reported experiences of teasing, with corresponding means (M) and standard deviations (SD), on validated measures of language, reading, and speech, in addition to subjective satisfaction with speech.

	No	Yes	
	% (n)	**% (n)**	
Reported teasing (parent reports)	65.8 (73)	34.2 (38)	
Language	**M (SD)**	**M (SD)**	**t**
Total Language score	93.4 (16.61)	91.8 (16.38)	.48
Reading			
Reading Comprehension	5.1 (1.79)	4.5 (2.11)	1.27
Word Chain Test	6.0 (1.90)	5.1 (1.88)	2.05*
Speech			
Resonance	.46 (.65)	.54 (.58)	-.52
Intelligibility	.34 (.52)	.35 (.56)	-.05
Subjective speech (self-reports)	8.6 (1.85)	7.4 (2.53)	2.53*

Note: * $p < .05$.