When there is more than a cleft: Psychosocial adjustment in children with an associated condition

Kristin Billaud Feragen

Bredtvet Resource Centre, Bredtvetveien 4, 0950 Oslo, Norway

Nicola Marie Stock

Centre for Appearance Research, University of the West of England, Bristol, UK

Corresponding author:

Kristin Billaud Feragen

Bredtvet Resource Centre

Tel. +47 22 90 28 85

Fax: +47 22 90 28 10

E-mail: kristin.feragen@statped.no

Acknowledgements: The work with this paper was financed by funds from the Norwegian ExtraFoundation for Health and Rehabilitation, project number 2011/0198.

Abstract

Background: In spite of studies reporting a relatively high frequency of additional conditions in children with a cleft lip and/or palate (CL/P), almost no research focuses on this clinically important subgroup. The objective of this study was to compare psychosocial adjustment in children with CL/P, with and without an additional condition.

Design: Cross-sectional data based on routine psychological assessments at age ten years, with comparisons to National reference groups.

Setting**:** Centralised treatment (Country).

Participants: 205 children with CL/P (participation rate: 80.1 %) from three consecutive birth cohorts.

Outcome measure**:** The Strengths and Difficulties Questionnaire (self-report and parent report), and the Child Experience Questionnaire (self-report).

Results: 81 children (39.5%) were identified as having at least one condition in addition to the cleft. These children reported significantly more psychosocial difficulties than children with a cleft alone. Differences between specific conditions were minor. Children with a cleft alone (*n* = 124) reported mean scores that were comparable to those reported by the reference group. There were no differences in adjustment between children with a visible vs. a non-visible cleft.

Conclusions: The present study highlights the need for research to be conducted on children with CL/P who have additional conditions, in order to provide better knowledge and clinical care for a potentially vulnerable subgroup of children and their parents.

Keywords: Cleft lip and palate, psychosocial adjustment, SDQ, learning difficulties, developmental delay

Introduction

A large number of studies have been conducted to assess the psychosocial impact of a cleft lip and/or palate (CL/P) on children and their families. Studies provide a varied and often inconsistent picture: while some studies report a wide range of psychosocial difficulties, lower self-perceptions, poorer educational attainment and dissatisfaction with appearance in children with CL/P, others have produced findings which are indicative of positive adjustment and processes of strengths (see review papers such as Turner et al, 1997; Rumsey and Harcourt 2004; Hunt et al., 2005; Feragen, 2012; Stock and Rumsey, in press). Variations in findings probably reflect multifaceted and complex processes in adjusting to a visible difference, but also to differing methodological approaches, such as a large variation in the use of concepts and measures, in addition to differences in sample characteristics (Rumsey and Stock, in press). One challenge, which is seldom specifically addressed within the present area of research, is the question of inclusion and exclusion criteria. It is well-established that a high proportion of children in cleft samples are at risk for associated developmental and cognitive problems (Milerad et al. 1997; Swanenburg, V et al. 2003; Hyman et al. 2005). However, few studies if any have focused specifically on these subgroups. The first consequence of this may be that subgroups of children with additional conditions are included in CL/P samples. As a consequence, underlying cognitive difficulties may be a mediating or confounding variable in the design of some studies (Pope and Snyder, 2005, Feragen and Stock, submitted). The second consequence is a lack of knowledge about these excluded groups; this is of primary clinical importance because of the number of children and families affected. While cleft services around the world currently offer treatment and support to all children affected by CL/P, further research is needed to identify subgroups of children who may be at risk, to measure the impact of this on the child and the family and to guide appropriate treatment and care for this specific group of children.

Children with a cleft and their families face several challenges associated with the psychological and/or psychosocial impact of growing up with a facial difference, in addition to the burden of treatment (Baker et al., 2009; Feragen, 2012; Rumsey and Stock, in press). If the child has one or more additional condition which also affects the family's and the child's daily life, it could present an additional burden of being different, and may imply a need for differing or specific treatment requirements. Higher levels of psychological difficulties such as depression are well documented in groups of individuals with learning difficulties (Huntington and Bender, 1993; Svetaz et al., 2000) and in children with chronic conditions, including CL/P (Pinquart and Shen, 2010; Martinez et al., 2011). Children with several additional conditions seem to be more vulnerable to psychological and emotional stress (Pinquart and Shen, 2011). In addition, research seems to indicate that children who look different may experience more teasing (Turner et al, 1997), a powerful negative social experience that could also be expected to be reinforced in children with a cleft and an additional condition. Children who are born with a cleft lip, compared to those with a cleft palate only, will also have to deal with the visibility of their condition, a difference that might reinforce the burden of an associated condition. One could therefore assume that the presence of a co-occurring condition could put children at risk for more adjustment difficulties compared to children with a cleft only.

The aim of the current study was to assess psychological, emotional and social strengths and difficulties in a representative sample of children with CL/P; a sample that included children with and without any other co-occurring conditions. The study explored the question of psychosocial adjustment with two perspectives in mind: first, to assess whether children with difficulties in addition to a cleft were at higher risk for psychological distress than children with a cleft but without any other associated conditions, and second, to assess whether subgroups of children with specific conditions were more at risk than others. Since some studies suggest that children who are visibly different may experience more teasing, potential differences between children with visible vs. non-visible clefts were examined. In addition, SDQ norms (www.sdqinfo.com) and related literature (Van Roy et al., 2010) refer to differences in self-reports and parent reports. The research questions were therefore explored from both perspectives in the current study. Scores on both psychological measures were compared to those provided by children from a large national comparison group.

Method

Participants and data collection

The present study was based on retrospective clinical data from psychological assessments at age ten. Data was collected from children with CL/P undergoing treatment by one of the two teams in (Country), a country providing centralised treatment of CL/P. With a 98–99% turnout for psychological examinations, the team has very few problems with non-attendance. Three consecutive birth cohorts of children born between 2000-2002, in addition to half of the birth cohort of children born in 1999 (*n* = 273) were included in the study. Fourteen children did not go through the routine psychological assessment because of severe developmental difficulties. Another three children did not complete the main outcome variable because of time constraints or non-availability of a psychologist on the day the family attended the clinic. The remaining 256 families were informed about the study and asked to participate. Participation rate was 80.1 % (*n* = 205/256). The sample included 132 children with CLP or CLA (64.4 %) and 73 children with CP/sCP (35.6 %).

The study conformed to guidelines by the local ethics committee (Regional Committee for Medical Research Ethics, Region (City)-East) and was hence based on informed consent from the child's parents. Information about the study was also sent specifically to the child.

Measures

Additional conditions and difficulties

Information about the presence of an associated condition was collected from the child's clinical records and/or from information provided by the parents at the time of assessment.

Associated difficulties affecting cognitive and/or psychological function included a wide range of conditions, such as a syndrome with or without other associated conditions (*n* = 16, 7.8 %), developmental difficulties (such as a diagnosis within the autism spectrum disorder, a developmental delay or more unspecific developmental difficulties affecting the child's cognitive capacities and learning, *n* = 22, 10.7 %), attention and/or hyperactivity disorders (AD/HD, *n* = 25, 12.2 %), specific language impairment (SLI, *n* = 16, 7.8 %), dyslexia (*n* = 10, 4.9 %) or general learning difficulties (*n* = 23, 11.2 %). In total, 81 children (39.5 %) had one or several conditions in addition to the cleft. The number of children with a specific condition does not add up to the total number of children with an additional condition since some children had more than one condition (*n* = 29).

Strengths and Difficulties Questionnaire (*SDQ*)

The SDQ (Goodman, 1997) is a brief 25 item screening measure of psychological difficulties and strengths in children. The SDQ has been used systematically at psychological routine assessment at age ten in the (City)-team since mid-2009, which means that data exists for more than three consecutive birth cohorts. The SDQ was completed by the child and one of the parents. The SDQ includes five subscales measuring emotional distress, conduct problems, hyperactivity/attention difficulties, peer relationship problems, and pro-social behavior. Each subscale includes five items that are positively or negatively worded. Each item is scored “not true”, “somewhat true”, and “certainly true” (0-2). The first four subscales are summarized into the Total difficulties score (including in total 20 items, with a total score ranging from 0-40). Standard cut-offs were used in order to identity children within the normal, borderline and high risk range (Goodman, 1997; Rønning et al., 2004). Internal reliability (Cronbach’s alpha) was satisfactory for the total 20 items score in both the child and the parent version of the questionnaire ($\alpha = .77$ and .84) and was either satisfactory or modest for some of the subscales: Emotional distress ($\alpha = .66$ and .65), Conduct problems ($\alpha = .48$ and .58), Hyperactivity/Attention difficulties ($\alpha = .58$ and .80), Peer relationship problems ($\alpha = .51$ and .64), and Pro-social behavior ($\alpha = .65$ and .62). Similar reliability have been reported in previous studies (Goodman, 2001; van Roy et al., 2008), and could mean that some of the subscales should be interpreted with caution. During the past years, the SDQ has been used in several population-based and clinical-based studies, and has been shown to be valuable screening instrument (Obel et al., 2004; Van Roy et al., 2006).

The Child Experience Questionnaire (CEQ)

Social experiences were measured by the Childhood Experience Questionnaire (CEQ, Pertschuk and Whitaker, 1982). The CEQ reflects the child's self-reporting of positive and negative social experiences on a five-point Likert scale. The questions in the scale relate to topics such as relations with friends ("I play with friends at school", "I have friends over to play"), social isolation ("I try to hide from people", "I keep to myself") and the perception of negative social experiences ("I am teased", "People stare at me"). Both positively and negatively worded items are included in the instrument, to avoid systematic response bias. Scores are converted so that high scores on the CEQ reflect positive social experiences. A mean total score was calculated (0-4), where four represented positive social experiences. The scale has been shown to possess satisfactory internal consistency and a coherent factor structure (Emerson et al., 2004). In the current study, the CEQ showed acceptable internal consistency reliability, with a Cronbach's $\alpha = .71$ for the 20 items.

Statistical Method

Analyses were performed using SPSS 19. Chi-square tests were performed when analysing differences in frequency between dichotomous groups, while differences between groups on the continuous outcome measures were analysed using ANOVA or independent sample *t*-tests. Cohen's *d* effect sizes were calculated in cases of significant differences, by using the means and standard deviations. Standard cut-offs (www.sdqinfo.com) were used when exploring the frequency of children scoring within the normal, borderline, or high risk groups on the SDQ (set by the following bandings; 80 % of the children in a sample within the normal range, 10 % borderline, and 10 % in the high risk clinical range). Cross-informant agreement (child vs. parent) was determined by using Pearson's correlation coefficient and paired samples *t*-tests (child vs. parent pairs). Effect sizes also calculated the magnitude of the differences in the child-parents pairs, correcting for the dependence between means.

Results

The Child Experience Questionnaire (CEQ)

Data from the Child Experience Questionnaire (CEQ) were missing for 47 children in the sample. Total mean scores (*SD*) for the CEQ were 2.80 (.35) for the group of children with a cleft alone, and 2.56 (.42) for the children with an additional condition *(t (156) = 3.87, p < .001, ES = .62)*, indicating that children with a cleft alone reported significantly more positive social experiences than the children with a condition in addition to a cleft.

Mean scores for the CEQ were calculated separately for each of the 20 items, in order to investigate whether some specific items could reflect differences that would be clinically meaningful. Eight items significantly differentiated between the two groups, with effect sizes ranging from .35 to .55. Only two items had an effect size above .50 indicating a difference that could be clinically significant: "I play with friends at school" (*F* (154) = 12.20, *p* < .001, ES = .55) and "I am teased" *F* (157) = 12.20, *p* < .001, ES = .53).[1]

The Strengths and Difficulties Questionnaire (SDQ)

Data from the Strengths and Difficulties Questionnaire were missing for 23 parents and 27 children. Mean values were calculated for the Total Difficulties score and for each of the five subscales of the SDQ. No significant differences in means were found between informants when differentiating between whether the SDQ was completed by the mother (*n* = 92), the father (*n* = 39) or both parents together (*n* = 46). Hence, further analyses did not take this variable into account. Table 1 presents means (SD) of the self-report and the parents' SDQ scores for children with CL/P with or without an additional condition and for the reference group, which consisted of 8,154 children from the "normal population" between the ages of 10-13 years and their parents (Van Roy et al., 2010)[2]. As can be seen in Table 1, the mean scores for children with a cleft alone were within the normal range for all subscales and similar to mean scores from the reference group. In contrast, children with a cleft and some additional condition had significantly higher mean scores on all scales of the SDQ, except

---

[1] The other differentiating items were: "I get into fights", *F* (155) = 5.47, *p* < .05, ES = .36); "I am chosen first", *F* (150) = 7.89, *p* < .01, ES = .47); "After school I spend a lot of time in front of the television", *F* (155) = 4.82, *p* < .05, ES = .35); "I am trying to hide myself", *F* (155) = 8.29, *p* < .01, ES = .44); "I am chosen last", *F* (149) = 5.52, *p* < .05, ES = .38); "I keep on by myself", *F* (155) = 6.62, *p* < .05, ES = .41).

[2] Mean scores from the reference group were presented separately for boys and girls in the current reference and were summed up when used in Table 1.

for the Pro-social behavior subscale. Mean scores were within the upper normal to borderline range, indicating a higher risk for psychological and emotional symptoms. Effect sizes (Cohen's *d*) are reported in Table 1 and demonstrated moderate (0.50 – 0.80) to large (from 0.80) effect sizes on all subscales except for self-reported Conduct difficulties (ES = 0.44) and Peer problems (0.46), with lower effect sizes.

Mean scores were also calculated for subgroups of children to investigate whether children with specific additional conditions (developmental difficulties or delay, AD/HD, SLI, specific learning difficulties or dyslexia) were at risk for psychological and/or emotional distress as measured by the SDQ. All subgroups had similar scores to those presented in Table 1 for children with an additional condition. In contrast, children with dyslexia ($n = 10$) differed from the other subgroups; mean scores fell within the clinical range, indicating a risk for psychological and emotional difficulties. Independent sample *t*-tests were performed on this data and indicated that several subscales significantly differed from scores of children with other additional conditions. On self-reports, mean scores (SD) for the children with dyslexia (M1) and children with some other additional condition (M2), *t*-test results, and effect sizes (ES, when significant *p*) were as follows: Total score: M1 = 18.5 (5.15), M2 = 13.5 (5.33), *t (63) = 2.50, p < .05, ES = .95*; Emotional difficulties: M1 = 5.5 (2.56), M2 = 3.7 (2.41), t (63) = 1.96, *p* = .055, ES = .72; Conduct difficulties: M1 = 3.1 (2.03), M2 = 2.1 (1.63), t (63) = 1.64, *p* > .05; Attention difficulties: M1 = 5.9 (1.46), M2 = 5.2 (2.05), t (63) = 0.89, *p* > .05; Peer difficulties: M1 = 4.0 (2.27), M2 = 2.5 (1.91), t (63) = 2.04, *p* < .05, ES = .72; Pro-social behavior: M1 = 7.0 (2.51), M2 = 8.1 (1.74), t (63) = -1.64, *p* > .05. For parent-reports, mean scores (SD), *t*-test results, and ES were as follow: Total score: M1 = 16.9 (7.00), M2 = 11.8 (6.64), t (66) = 2.01, *p* < .05, ES = .75; Emotional difficulties: M1 = 3.4 (2.62), M2 = 2.8 (2.21), t (66) = .69, *p* > .05; Conduct difficulties: M1 = 3.0 (1.60), M2 = 1.7 (1.72), t (66) = 2.04, *p* < .05, ES = .79; Attention difficulties: M1 = 7.3 (2.60), M2 = 4.8 (2.90), t (66) = 2.25, *p* < .05, ES = .91; Peer difficulties: M1 = 3.3 (2.19), M2 = 2.5 (2.29), t (66) = .83, *p* > .05; Pro-social behavior: M1 = 8.3 (1.91), M2 = 8.7 (1.53), t (66) = -.77, *p* > .05. As can be seen, all calculated effect sizes were large.

According to SDQ cut-off scores (Van Roy et al., 2006 and www.sdwinfo.com), the reported means place the children with dyslexia at particularly high risk for psychological or emotional distress. Parent reports indicated scores within the borderline range for conduct problems, high risk range for attention difficulties and borderline/high risk range for peer problems. Self-reports indicated borderline/high risk for emotional distress, normal/borderline risk for attention difficulties and borderline risk for peer problems.

Frequency of children scoring within the normal vs. clinical range

Cut-off scores (Goodman, 1997; Van Roy et al., 2006) have been set to identify children scoring within the normal, borderline and clinical range. Table 2 shows that the prevalence of children with a cleft alone scoring within the normal range was similar to the normal population. In contrast, a substantially higher group of children with CL/P and an additional condition were likely to score within the clinical ranges of the SDQ. According to self-reports, approximately 12 % of the children with a cleft alone had scores within the borderline/high risk range, compared to 20 % in the reference group, while approximately 45 % of children with a cleft and an additional condition were at risk for psychological and emotional difficulties ($\chi^2 = 22.44$, $p < .001$). The difference in frequency of risk according to self-reports was also present on the following subscales: Emotional difficulties ($\chi^2 = 7.34$, $p < .01$), Conduct difficulties ($\chi^2 = 8.24$, $p < .01$), Attention difficulties ($\chi^2 = 11.47$, $p < .01$) and Peer problems ($\chi^2 = 3.10$, $p = .059$), while there were no reported differences in Pro-social behavior ($\chi^2 = 1.12$, $p > .05$). Parent reports confirmed the same findings, summarized by the Total score ($\chi^2 = 36.91$, $p < .001$) and in the following subscales: Emotional difficulties ($\chi^2 = 12.00$, $p < .01$), Conduct difficulties ($\chi^2 = 5.89$, $p < .05$), Attention difficulties ($\chi^2 = 24.71$, $p < .001$) and Peer problems ($\chi^2 = 24.73$, $p < .001$). As for self-reports, no differences were found in relation to Pro-social behavior ($\chi^2 = 1.00$, $p > .05$).

Cleft visibility

As can be seen in Tables 3 and 4, no differences were found between children with a non-visible vs. a visible cleft on self-reports or on parent reports. This was the case for children with a cleft only, and children with a cleft and an additional condition.

Agreement and differences between children and parent scores

The agreement between self-reports and parent scores on the SDQ's Total score was $r = .46$ ($p < .01$). On the subscales, the sizes of correlation coefficients were moderate and varied from $r = 0.26$ (Pro-social behavior) to $r = 0.49$ (Peer problems). All correlations were significant at the .01 level and comparable or somewhat higher than what has been reported from large national reference groups (Van Roy et al., 2010).

Paired sample *t*-tests showed that children reported more symptoms than their parents on all subscales. Total score: t (170) = 6.33, $p < .001$, ES = .49; Emotional difficulties: t (171) = 5.33, $p < .001$, ES = .41; Conduct difficulties: t (171) = 3.05, $p < .01$, ES = .23; Attention difficulties: t (171) = 4.43, $p < .001$, ES = .35; Peer difficulties: t (171) = 4.58, $p < .001$, ES = .35; Pro-social behavior: t (171) = -1.70, $p < .01$, ES = -.21. As can be seen, all differences in means between child and parent scores were significant. However, effect size calculations revealed that these differences were small for all subscales, while being small to moderate for the Total score.

Discussion

The aim of the present study was explore psychological, emotional and social strengths and difficulties in a representative sample of children with a cleft, with and without any other co-occurring conditions. The results indicated that children with a cleft and an additional condition were at increased risk for psychological and emotional problems, and also reported less positive social experiences, while children with a cleft alone reported adjustment scores that were within the normal range, and similar to the comparison group. Cut-off scores classifying children within the normal or clinical range demonstrated that a significantly higher proportion of the children with a cleft and an additional condition had scores within the borderline/high risk range, compared to the children with a cleft only and to the reference group. Findings were supported by both child and parent reports. Analyses across conditions seemed to indicate that the most useful classification is whether there is, or whether there is not, a co-morbid condition in addition to the cleft. However, among the children with an additional condition, one subgroup emerged: children with dyslexia appeared to be more at risk than children with other additional conditions. There were no differences between children with a visible vs. a non-visible cleft, irrespective of whether the child had an additional condition or not.

When there is more than a cleft

The findings of the present study clearly identified children with a cleft and an additional difficulty as a risk group, while children with a cleft alone had adjustment scores within the normal range, and comparable to the reference group. While psychosocial adjustment in the group of children with a cleft only were in line with a similar study using some of the same measures (Berger and Dalton, 2009), no studies have, to the authors' knowledge, specifically addressed the question of psychosocial adjustment in children with a cleft and an associated condition. However, two recent studies (Baker et al., 2009; Stock and Rumsey, in preparation/2012) mentioned that the presence of a medical condition in addition to the cleft may impact significantly on the family, in concordance with the results of the current study. To our knowledge, this is the first study to investigate psychosocial adjustment in children with CL/P who also have to cope with a co-occurring condition. Results clearly indicate that the presence of an additional condition was perceived, both by children and parents, to have impacted on a range of psychological and emotional issues, compared to children with a cleft only and to children from the reference group. These results are in line with previous findings (Feragen and Stock, in submission) and suggest the importance of careful categorisation of co-occurring conditions, as well as highlighting a need for further knowledge about this potentially more vulnerable subgroup.

The results of the present study correspond to previous research and meta-analyses which show children with a chronic condition to be at higher risk for psychological, emotional and/or social distress (Lavigne and Faier-Routman, 1992; Martinez et al., 2011; Pinquart and Shen, 2011; Ferro and Boyle, 2012). We asked whether having an extra condition in addition to a visible cleft, as opposed to an additional condition associated with a non-visible cleft, would represent an increased vulnerability. Studies have reported a higher frequency of teasing and appearance-related bullying in children with a visible difference such as a cleft (Turner et al., 1997; Hunt et al, 2006), and among children with other problems, such as reading difficulties (Terras et al., 2009). Hence, the group of children with a visible cleft and some additional learning difficulties could be expected to be particularly vulnerable. However, the results of this study did not indicate differences in psychosocial adjustment between children with visible and non-visible clefts. The findings from the present study indicate that variables other than objective visibility account for the variation in psychological and emotional distress; a finding that is reasonably well-documented in psychological research investigating the impact of a visible difference (Rumsey and Harcourt, 2004; Feragen, 2012). This could be in accordance with a study demonstrating that the association between appearance dissatisfaction and the visibility of the cleft was mediated by experiences of peer harassment (Feragen and Borge, 2010), which highlights the importance of the child's social experiences for psychological health and well-being, irrespective of the visibility of a condition.

Results from the present study indicated that children with an additional condition and their parents reported higher levels of teasing and negative social experiences than children with a cleft only, as was demonstrated by the findings on the CEQ and the SDQ. The CEQ specifically pointed to differences in the child's perception of social experiences: children with an associated condition reported a lower frequency of playtime with friends, and more time spent on activities such as watching television or keeping on by themselves. In line with findings from the CEQ, self-reports on the peer problem subscale of the SDQ indicated more social difficulties in the group of children with an associated condition. Negative social experiences could therefore partly explain the higher frequency of psychological and emotional distress in children with an additional condition such as a learning difficulty, as was found in the current study. Parent reports on the peer problem subscale seem to confirm the child's self-reports on the SDQ. However, the present study cannot answer the question of whether reported and/or experienced teasing is a cause or an effect of psychosocial vulnerability. It is also important to remember that parents of children with challenging conditions, or conditions they experience as challenging, could be more susceptible to perceiving and interpreting ambiguous social experiences as negative, and to developing perceptions of child vulnerability (Noll and Bukowski, 2012). We know that expectations affect perceptions and interpretations of social interactions (Thompson and Kent, 2001; Feragen et al., 2009). Further, perceptions of negative experiences may contribute to the development of negative self-appraisals, as well as to social isolation and emotional distress. Such processes can be expected to be at work in the child as well as in their parents. As a consequence, factors influencing parent's and children's cognitions should be clarified, and perceptions of child vulnerability should also be directly explored (Anthony et al., 2003).

Children with CL/P and dyslexia

Among the subgroups of children with additional conditions, children with dyslexia seemed particularly at risk for emotional and psychological distress. This is in line with previous research indicating that children with dyslexia may be a psychologically vulnerable group (for a review, see Maughan and Carroll, 2006). Results from the current study were similar to those reported in a study on children with dyslexia which also used the SDQ (Terras et al., 2009), confirming that this could be a more vulnerable group irrespective of whether the child has CL/P or not. Given the small number of children in this subgroup, the present findings would need to be replicated and checked in larger samples; to explore whether the findings were associated with specific characteristics within this particular group of children, or whether the study identified a particularly vulnerable subgroup which CL/P clinicians should be aware of. In both cases, more research is needed in order to enhance our knowledge of potentially vulnerable subgroups in CL/P samples, so that these children and their families are identified early on and treated appropriately.

The results of this study are believed to be important for the long-term care provided by the cleft-teams worldwide, and to the information given to parents regarding how their child is likely to cope with the additional challenges he/she was born into. If a broad categorization providing details of co-occurring conditions is clinically valid, as the findings of the current paper suggest, this categorization would provide professionals and clinicians with more adequate tools when informing new parents about their child's future adjustment.

Methodological issues

Internal reliability calculations showed that some subscales of the SDQ should be interpreted with caution. On self-reports, the Total score had satisfactory values of internal reliability, while the Conduct and Peer problems subscales demonstrated questionable internal reliability. Parent reports demonstrated less problematic internal consistency, but values were still questionable on some of the subscales. However, findings from the current study were in line with reports from large national samples (Rønning et al., 2004, van Roy et al., 2008), and with internal consistency reports by Goodman (2001). In spite of potentially problematic subscales, it appears that the Total score of the SDQ provides an accurate picture (Richter et al., 2011). This also strengthens the findings from the present study, since reliability and effect sizes were large in the current CL/P sample.

Children reported more distress than their parents, as has been reported in the reference groups (Van Roy et al., 2006; Van Roy et al., 2010). In general, agreement between children and parents was comparable to reports in previous large-scale studies (Van Roy et al. 2010). Some subscales, such as the Pro-social subscale, showed lower cross-informant agreement, while parents and children seemed to agree more on the presence or absence of social and peer problems. However, it is important to remember that lower agreement does not necessarily reflect a lack of valid reports from one of the informants, but can be explained by a uniquely different perception of challenges and strengths. High scores from one of the informants on some subscale could probably point to areas that the informant experience as difficult or challenging, areas that hence should be explored clinically.

Previous studies have shown a need for a larger consensus in the use of concepts and measures in cleft research (Rumsey and Stock, in press). The present study shows that the SDQ could be a potentially useful screening tool for identifying children and adolescents at risk in cleft care: It can be completed in a short time, it is easily available in downloadable versions along with scoring instructions and norms, and it has been translated into many different languages. Hence, the SDQ could prove itself to be a good and useful measure for use in cleft clinics, and provide opportunities for easier comparisons between studies and across samples.

Strengths and limitations

Several limitations and strengths of this study should be addressed. One strength of the study was the high participation rate, with data collected from within a centralised treatment setting; this suggests that the sample is highly representative. Another strength was the use of a well-known and valid questionnaire, followed by the question of whether the SDQ could prove itself to be a valuable measure in cleft research. Third, the outcome measure was provided by both the parents and the child, strengthening the findings and the study's methodology. The participation of fathers, who often are not represented in the present area of research (Stock, and Rumsey, in preparation), was also a strength. The use of two different measures were also a strength in the study, since the CEQ provided some additional information on the child's subjective experience of his/her social experiences. Nonetheless, some caution is required when interpreting the results. The CEQ has been criticised for a somewhat unclear factor structure (Emerson et al., 2004), and reliability measures were no more than acceptable. However, compared to reliability in the SDQ, internal consistency could be viewed as satisfactory. Further, the SDQ was constructed for use with children from the age of eleven years and above, while the children in the present study were aged ten at the time of assessment. However, children of the same age were included in the large national study which has been used as a reference group to the present results (van Roy et al., 2010). A third limitation is the relatively low reliability measures for some of the subscales. This could indicate that some children have problems with understanding and using the measure, which may be due to their age. However, methodological findings were again very similar to those reported by Goodman (2001) and in national samples (Van Roy et al., 2008). A fourth limitation of the present study is the lack of a control group. However, it was possible to compare this study's findings with results from national samples and scale norms. A final limitation is associated with variables that were not included and hence not controlled for in the present study, such as speech difficulties, that could be expected to be more frequent in children with some associated conditions (Ref…???). Children that are difficult to understand could be among the ones reporting social isolation and/or teasing. Future studies should include information about speech in order to address this question.

Conclusion

The main implication of the present study is the need to recognise the increased risk of psychological and emotional difficulties in children who have difficulties in addition to a cleft.

The results appear to be even stronger when taking into account that fourteen children were excluded from the study due to severe developmental difficulties, which impeding a standardised routine assessment and the use of the outcome measures. In order to reduce the burden of assessments for these parents, questionnaires are usually replaced by a conversation. If parent reports from the SDQ had been included for this group, the results may indicate an even higher risk. Given the results of this study, an important direction for future research would be to move beyond the broad question of psychosocial adjustment to CL/P, and towards the exploration of subgroups within this population more specifically. In order to be able to do so, large and representative samples are needed.

Further research is needed in order to identify vulnerability factors, and factors which promote positive adjustment in children with a cleft. We need to identify the specific risk factors at work in this group of children which increase the likelihood of problematic adjustment, rather than entailing negative outcomes in the group as a whole. The identification of whether there are additional conditions or not could be one such major factor. Although the results of the present study suggest that children with an additional condition are at greater risk for psychological distress as a group, it is important to remember that there are significant individual differences in response to the challenges they face. There is a need for a better understanding of factors associated with risk or resilience in order to be able to promote positive outcomes for these children. The results of the current study indicate that children with an additional condition are at risk for psychosocial and emotional difficulties, whereas those with a cleft alone score within the normal range, indicating good psychological and emotional adjustment. In addition, the results highlight the need for more research with children who have a condition in addition to CL/P, and the need for early identification and intervention.

References

Anthony, K.K, Gil, K.M., Schanberg, L.E. Brief report: Parental perceptions of child vulnerability in children with chronic illness. *J Pediatr Psychol.,* 2003; 28: 185-190.

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

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

Emerson, M., Spencer-Bowdage, S., & Bates, A. Relationships between self-esteem, social experiences and satisfaction with appearance: standardisation and construct validation of two cleft audit measures. In The Craniofacial Society of Great Britain and Ireland. Annual Scientific Conference. "Setting a positive agenda in cleft and craniofacial care", April 2004, Bath, United Kingdom.

Feragen, K.J.B. (2012). Experiences of people living with visible differences. Congenital conditions. In N. Rumsey & D. Harcourt (Eds). *Oxford Handbook of Psychology of Appearance,* Oxford University Press, 2012: 353-371.

Feragen, K.J.B., Borge, A.I.H. Peer harassment and satisfaction with appearance in children with and without a facial difference. *Body Image,* 2010; *7: 97-105.*

Feragen, K. J. B., Borge, A. I. H., & Rumsey, N. Social experience in 10-year-old children born with a cleft: Exploring psychosocial resilience. *Cleft Palate Craniofac J.,* 2009; *46:* 65-74.

Feragen, K.J.B., Stock, N. Towards a reconsideration of inclusion and exclusion criteria in cleft lip and palate: Implications for psychological research. Submitted to *Cleft Palate Craniofac J.,* Submitted to CPCJ.

Ferro, M.A., Boyle, M.H. Longitudinal invariance of measurement and structure of global self-concept: A population-based study examining trajectories among adolescents with and without chronic illness. *J Pediatr Psychol.*, 2012: 1–13.

Galambos, N.L., Leadbeater, B.J., & Barker, E.T. Gender differences in and risk factors for depression in adolescence: A 4-year longitudinal study. *Int J Beh Dev,* 2004; 28: 16-25.

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

Goodman, R. Psychometric properties of the Strengths and Difficulties Questionnaire, *J Am Acad Child Adolesc Psychiatry*, 2001; 40: 1337-45.

Hankin, B.L., Abrahamson, L.Y. Development of gender differences in depression: Description and possible explanations. *Ann Med.,* 1999; 31: 372-379.

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

Huntington, D. D., & Bender, W. N. Adolescents with learning disabilities at risk?
Emotional well-being, depression, suicide. *J Learn Disabil.*, 1993; 26: 159–166.

Hyman, S.L., Shores, A., and North, K.N. The nature and frequency of cognitive deficits in children with neurofibromatosis type 1. *Neurology*, 2005; 65: 1037-1044.

Lavigne, J.V. & Faier-Routman, J. Psychological adjustment to pediatric physical disorders: A meta-analytic review. *J Pediatr Psychol.*, 1992; 17: 133-157.

Martinez, M., Smith Carter, J., Legato, L.J. Social competence in children with chronic illness: A meta-analytic review. *J Pediatr Psychol.,* 2011; 36: 878-890.

Maughan, B., & Carroll, J. Literacy and mental disorders. *Curr Opin Psychiatry*, 2006; 19: 350–354.

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

Noll, R.B., Bukowski, W.B. Commentary: Social competence in children with chronic illness: The devil is in the details. *J Pediatr Psychol.,* 2012; 37: 959-966.

Obel, C., Heiervang, E., Rodriguez, A., Heyerdahl, S., Smedje, H., Sourander, A., Gudmundsson, O.O. et al. The Strengths and Difficulties Questionnaire in the Nordic countries. *Eur Child Adolesc Psychiatry*, 2004; 13: 32-39.

Parker, G. & Brotchie, H. (2010). Gender differences in depression. *Int Rev Psychiatry,* 2010; 22: 429-436.

Pertschuk, M. J. & Whitaker, L. A. Social and psychological effects of craniofacial deformity and surgical reconstruction. *Clin Plast Surg.*, 1982; 9: 297-306.

Pinquart, M., Shen, Y. Behavior problems in children and adolescents with chronic physical illness: A meta-analysis. *J Pediatr Psychol.,* 2011; 36: 1003-1016.

Pope, A.W., Snyder, H.T. Psychosocial adjustment in children and adolescents with a craniofacial anomaly: Age and sex patterns. *Cleft Palate Craniofac J.*, 2005; 42: 349-354.

Richter, J., Sagatun, Å., Heyerdahl, S., Oppedal, B., Røysamb, E. The Strengths and Difficulties Questionnaire (SDQ) - self-report. An analysis of its structure in a multiethnic urban adolescent sample. *J Child Psychol Psdychiatry,* 2011; 52:1002-1011.

Rønning, J.A., Handegaard, B.H., Sourander, A., Mørch, W.-T. The Strengths and Difficulties self-report Questionnaire as a screening instrument in Norwegian community samples. *Eur Child Adol Psychiatry,* 2004; 13: 73-82.

Rumsey, N., Harcourt, D. Body image and disfigurement: issues and interventions. *Body image*, 2004; 1: 83-97.

Rumsey, N. & Stock, N.M. (in press). Living with a cleft: Psychological challenges, support and intervention. In: S. Berkowitz (Ed.), Cleft Lip and Palate: Diagnosis and Management (third edition). Springer-Verlag Berlin Heidelberg.

Stock, N.M. & Rumsey, N. (in preparation). Parenting a child with a cleft: The father's perspective. Centre for Appearance Research, UWE, Bristol.

Svetaz, M.V., Ireland, M., Blum, R. Adolescents with learning disabilities: risk and protective factors associated with emotional well-being: findings from the National Longitudinal Study of Adolescent Health. *J Adol Health*, 2000; 27: 340-348.

Swanenburg, d., V, Beemer, F.A., Mellenbergh, G.J., Wolters, W.H., and Heineman-de Boer, J.A. An investigation of the relationship between associated congenital malformations and the mental and psychomotor development of children with clefts. *Cleft Palate Craniofac J.*, 2003; 40: 297-303.

Terras, M.M., Thompson, L.C., Minnis, H. Dyslexia and psychosocial functioning: An exploratory study of the role of self-esteem and understanding. *Dyslexia*, 2009; 15: 304–327.

Thompson, A., Kent, G. Adjusting to disfigurement processes involved in dealing with being visibly different. *Clin Psychol Review*, 2001; 21: 663-682.

Turner, S.R., Thomas, P.W., Dowell, T., Rumsey, N., and Sandy, J.R. Psychological outcomes amongst cleft patients and their families. *Br J Plast Surg*, 1997; 50: 1-9.

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

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

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

Wyszynski, D.F., Sárközi, A., Czeizel, A.E. Oral clefts with associated anomalies: Methodological issues. *Cleft Palate Craniofac J*, 2006; 43: 1-6.

TABLE 1. Psychological and emotional adjustment in children with a cleft as measured by the SDQ: self-reports (*n* = 173) and parent reports (*n* = 177).

	Reference group	**Cleft only**	**Cleft and additional condition(s)**	Tests of significance	
Self-report	M (SD)	*M* (*SD*)	*M* (*SD*)	*t*	*ES*
Total score	10.2 (5.15)	9.6 (4.56)	14.1 (5.52)	-5.88***	0.89
Emotional difficulties	2.6 (2.15)	2.6 (2.07)	3.9 (2.48)	-3.87***	0.57
Conduct difficulties	1.9 (1.55)	1.5 (1.45)	2.2 (1.71)	-3.01**	0.44
Hyperactivity/inattention	3.7 (2.15)	3.8 (2.00)	5.3 (1.99)	-4.85***	0.75
Peer problems	2.0 (1.75)	1.9 (1.69)	2.7 (2.00)	-2.75**	0.43
Prosocial behaviour	7.8 (1.70)	8.3 (1.84)	8.0 (1.86)	.90	n/a
Parent reports					
Total score	6.2 (5.00)	5.8 (4.25)	12.4 (6.82)	-7.27***	1.16
Emotional difficulties	1.3 (1.75)	1.5 (1.68)	2.9 (2.25)	-4.22***	0.71
Conduct difficulties	1.1 (1.30)	1.1 (1.26)	1.8 (1.75)	-3.29**	0.46
Hyperactivity/inattention	2.6 (2.20)	2.4 (2.05)	5.1 (2.96)	-6.74***	1.06
Peer problems	1.2 (1.65)	.9 (1.34)	2.6 (2.28)	-5.73***	0.91
Prosocial behaviour	8.3 (1.60)	8.6 (1.64)	8.7 (1.57)	-.43	n/a

Reference group from van Roy, Grøholt, Heyerdahl, & Clench-Aas, 2010.

Independent sample *t*-test and effect sizes (ES) between the two cleft groups (with and without an additional condition).

*$p< .05$; **$p < .01$; ***$p< .001$.

TABLE 2. Frequency of children with a cleft with and without an additional condition.

	Cleft only			**Cleft with additional condition(s)**		
	Normal % (*n*)	Borderline % (*n*)	High risk % (*n*)	Normal % (*n*)	Borderline % (*n*)	High risk % (*n*)
Self-report						
Total score	87.9 (94)	5.6 (6)	6.5 (7)	56.9 (37)	16.7 (11)	27.3 (18)
Emotional difficulties	83.2 (89)	10.3 (11)	6.5 (7)	65.2 (43)	9.1 (6)	25.8 (17)
Conduct difficulties	93.5 (100)	1.9 (2)	4.7 (5)	78.5 (51)	10.6 (7)	10.6 (7)
Hyperactivity/inattention	81.3 (87)	10.3 (11)	8.4 (9)	57.6 (38)	16.7 (11)	25.8 (17)
Peer problems	81.3 (87)	9.3 (10)	9.3 (10)	69.7 (46)	9.1 (6)	21.2 (14)
Prosocial behaviour	92.5 (99)	4.7 (50	2.8 (3)	87.9 (58)	6.1 (4)	6.1 (4)
Parent reports						
Total score	93.5 (101)	5.6 (6)	0.9 (1)	55.1 (38)	15.9 (11)	29.0 (20)
Emotional difficulties	88.1 (96)	8.3 (9)	3.7 (4)	66.7 (46)	10.1 (7)	23.2 (16)
Conduct difficulties	88.1 (96)	7.3 (8)	4.6 (5)	73.9 (51)	8.7 (6)	17.4 (12)
Hyperactivity/inattention	89.0 (97)	7.3 (8)	3.7 (4)	56.5 (39)	10.1 (7)	33.3 (23)
Peer problems	88.1 (96)	5.5 (6)	6.4 (7)	55.1 (38)	13.0 (9)	31.9 (22)
Prosocial behaviour	96.3 (105)	2.8 (3)	0.9 (1)	95.7 (66)	1.4 (1)	2.9 (2)

Note: Cut-off points define a 10 % high risk groups and a 10 % borderline group (www.sdqinfo.com).

Table 3 Psychological and emotional adjustment in children with a non-visible or a non-visible cleft without any associated condition as measured by the SDQ.

	Non-visible clefts		**Visible clefts**		Tests of significance	
Child measures	Mean	SD	Mean	SD	*t*-value	*p*
SDQ Total score	10.1	4.77	9.4	4.49	.73	>.05
Emotional	2.6	2.27	2.6	2.00	-.04	>.05
Conduct	1.4	1.41	1.5	1.47	-.35	>.05
Attention/Hyper	3.9	2.23	3.7	1.90	.54	>.05
Social	2.2	1.82	1.8	1.63	1.15	>.05
Prosocial	8.3	1.91	8.2	1.83	.19	>.05
Parent measures						
SDQ Total score	5.5	4.02	5.9	4.36	-.36	>.05
Emotional	1.8	2.11	1.4	1.47	.89	>.05
Conduct	0.8	0.98	1.2	1.34	-1.59	>.05
Attention/Hyper	2.1	1.83	2.5	2.14	-.97	>.05
Social	1.0	1.49	0.9	1.29	.39	>.05
Prosocial	8.3	2.02	8.7	1.46	-1.21	>.05

Table 4 Psychological and emotional adjustment in children with a visible or a non-visible cleft and an associated condition as measured by the SDQ.

	Non-visible clefts		**Visible clefts**		Tests of significance	
Child measures	Mean	SD	Mean	SD	*t*-value	*p*
SDQ Total score	13.9	5.51	14.3	5.62	-.24	>.05
Emotional	4.2	2.50	3.7	2.48	.73	>.05
Conduct	1.8	1.42	2.6	1.89	-1.73	>.05
Attention/Hyper	5.6	2.14	5.0	1.82	1.34	>.05
Social	2.3	1.80	3.0	2.14	-1.47	>.05
Prosocial	7.9	1.74	8.1	2.00	-.53	>.05
Parent measures						
SDQ Total score	11.8	7.30	12.9	6.44	-.65	>.05
Emotional	2.9	2.35	2.8	2.19	.07	>.05
Conduct	1.6	1.79	2.0	1.71	-.95	>.05
Attention/Hyper	5.1	3.12	5.1	2.86	-.10	>.05
Social	2.3	2.22	2.9	2.31	-1.16	>.05
Prosocial	9.0	1.02	8.4	1.89	1.81	>.05