

The Scandcleft randomised controlled trials: Parent's perceptions of appearance and treatment outcomes in their 5-year-olds with unilateral cleft lip and palate

Background and aim: Few studies have explored children's emotional and behavioural reactions to cleft surgery and treatment-related stress. The objective was to investigate parents' evaluations of appearance and treatment outcomes in their 5-year-old child with unilateral cleft lip and palate (UCLP), and their perceptions of how their child was coping with treatment, comparing this information with recorded postsurgical complications.

Design: Three parallel group randomised clinical trials were undertaken as an international multicenter study by 10 cleft teams in five countries: Denmark, Finland, Sweden, Norway and UK.

Methods: Three different surgical procedures for primary palatal repair were tested against a common procedure in the total cohort of 448 children born with a nonsyndromic UCLP. A total of 356 parents completed the Scandcleft Parent Questionnaire and 346 parents completed the Cleft Evaluation Profile.

Results: The results indicated that the majority of parents were satisfied with cleft-related features of their child's appearance. Further, most children coped well with treatment according to their parents. Nevertheless, 17.5% of the children showed minor or short-term reactions after treatment experiences, and 2% had major or lasting difficulties. There were no significant relationships between parent perceptions of treatment-related problems and the occurrence of post-surgical medical complications.

Conclusions: Most parents reported satisfaction with their child's appearance. However, treatment-related problems were described in some children, urging cleft centres to be aware of potential negative emotional and behavioural reactions to treatment in some young children, with a view to preventing the development of more severe treatment-related anxiety.

Key words: Parent satisfaction; cleft; appearance; treatment anxiety; treatment-related problems; coping.

INTRODUCTION

This paper is one of a series of reports of the Scandcleft Project, consisting of three concurrent randomised trials of primary surgery for infants born with complete unilateral cleft lip and palate (UCLP). The project was developed and executed by ten North European cleft teams: Århus /Copenhagen (Denmark), Helsinki (Finland), Bergen/Oslo (Norway), Gothenburg/Linköping/Stockholm (Sweden), Manchester/Belfast (UK)

One surgical protocol was defined to serve as a common method in each trial against which a local protocol were compared. The common surgical protocol was lip and soft palate closure at 3-4 months and hard palate closure at 12 months. Trial 1 compared this with only a variation in timing: hard palate repair at 36 months; Trial 2 with lip repair at 3-4 months followed by hard and soft palate closure at 12 months; and Trial 3 with lip and hard palate repair at 3-4 months and soft palate repair at 12 months. Recruitment of 448 infants took place over a 9-year period with high subsequent retention of participants in each trial. The present series of reports include primary outcomes of speech and dentofacial development at age 5, and perioperative and longer term secondary outcomes. Background information about the project can be found in Semb et al.'s introductory paper [1].

The present series of reports include primary outcomes of speech and dentofacial development at age 5, and perioperative and longer term secondary outcomes. This paper considers parent's perceptions of appearance and treatment outcomes in their 5-year-old child with cleft lip and palate

Children born with a cleft lip and palate (CLP) undergo a number of procedures and surgical interventions during their first years of life, which are carried out to correct

and improve oral function, speech, and facial appearance. Studies have quantified the burden of hospital care for children with CLP [2,3], and surgical treatment can be a frightening experience for a child [4]. Few, if any studies have, however, explored parents' perceptions of how their child cope with cleft surgery and treatment-related stress, while also comparing this information with registered postsurgical complications and surgical technique.

Perceptions of appearance-related outcomes of cleft treatment is another important area in cleft research. A range of studies have explored this issue as assessed by professionals, lay-persons, patients and parents [5,6,7,8,9,10,11]. Studies comparing parent, patients, and professionals' satisfaction with cleft-related features report contradictory findings (for a review, see [10,12]), which may partly be explained by different methodological approaches, in addition to a significant diversity in the choice of concepts and outcome measures [13]. Few studies have compared different surgical techniques using the same outcome measure, across a large sample of parents from different countries, highlighting potential cultural differences within the same design and methodology.

When investigating associations between psychological adjustment and appearance satisfaction, research has quite consistently shown that subjective patient centered measures should be attended to in preference to more "objective" evaluations by professionals [14,15,16]. Parent perspectives are also valuable, particularly in the child's early years, and are important to include when evaluating treatment outcomes of randomised trials such as the Scandcleft trials, if cleft teams are to fully understand the complexity of satisfactory treatment outcomes [17]. Therefore, parent reports

were included in the Scandcleft project, in addition to evaluations of surgical, orthodontic, and speech outcomes.

AIMS

The aims of this part of the study were to: a) Investigate parents' evaluations of appearance and other cleft-related features in their 5-year-old child with cleft lip and palate, and b) Explore parents' perceptions of their child's response to treatment, comparing this information with registered postsurgical complications.

MATERIALS AND METHODS

A total of 448 patients were included in the Scandcleft trials, three of whom were lost to follow-up. Parents of 356 children at age 5 years completed the Scandcleft Parent Questionnaire (Participation rate: 80%). There were 33.3% girls ($n = 119$) and 67% boys ($n = 237$).

A total of 356 parents of 5-year-olds with complete unilateral cleft lip and palate, treated in the Scandcleft trials by nine North-European cleft centers from the United Kingdom, Denmark, Sweden, Norway, and Finland participated in the present study. The tenth centre did not hand out the questionnaires for parents to complete. Of these, 346 completed the Cleft Evaluation Profile (Participation rate: 78%), and 356 completed the Scandcleft Parent Questionnaire (Participation rate: 80%). There were 33.4% girls ($n = 119$) and 66.6% boys ($n = 237$) in the total sample. Parents who completed the questionnaire were couples (55.1%; $n = 196$), in addition to 126 mothers (35.3%) and 14 fathers (3.9%). Other informants were foster parents or grandparents (1.2%, $n = 4$), while 16 respondents (4.5%) did not indicate their relationship with the child.

Measures

Cleft Evaluation Profile (CEP): The CEP originated from the Royal College of Surgeons Cleft Lip and Palate Audit Group [6]. The 8 item scale was used to assess parents' perceived satisfaction with features associated with cleft and its treatment, namely speech, hearing, lip, nose, teeth, bite, breathing, and profile. Respondents are asked to rate their satisfaction on a 7-point Likert scale, ranging from very satisfactory (1) to very unsatisfactory (7). A total mean score of parent satisfaction was computed (mean score for each item divided by the number of items). Scores below 4 on individual items have previously been labelled as satisfactory [6]. Noor and Musa [8] suggested a cut-off score of 32 on the total score (summing up the score for each item) as a measure of satisfaction with treatment. The CEP has been reported to possess satisfactory internal consistency, with a Cronbach $\alpha = .84$ for parents of adolescents [8]. Internal reliability was high also in the present dataset ($\alpha = .81$).

The Scandcleft Parent Questionnaire: A structured, self-administered questionnaire was designed by clinical psychologist Dr. Eileen Bradbury (Manchester, UK), and was translated into the four remaining languages. The questionnaire consisted of four sections, including a range of questions about parental responses to the cleft and the diagnosis, and their perception of their child's reactions and coping. Only parental reports of the child's responses and reactions to treatment are included in the present article: "How has your child coped with attending: a) Speech therapy, b) Surgery, and c) Other (Please specify)". One of the countries had an open-response format to this question without any mention of disciplines.

Procedure: The questionnaire was handed out by the teams' speech and language pathologists/therapists (SLP/T) when the families attended the child's 5-year-old

multidisciplinary assessment in four of the five participating countries. In one country, the questionnaire was sent by post to the parents prior to the 5-year-old assessment, and parents were asked to return it when attending the cleft centre. The parents were informed in the introductory paragraph of the questionnaire that their responses would be anonymised. Ethical consent was sought locally by each participating treatment centre and/or country.

Statistical analyses

Analyses were performed using SPSS 22 (IBM Corp, Armonk, NY). Means and standard deviations were calculated for the CEP, and compared across gender, trials and arms with ANOVA. Tukey multiple-comparison test was used when investigating differences between arms within trials. A total CEP score based on the sum of all means was calculated, so that cut-off scores could be applied [8].

Parents reports regarding their child's responses to treatment were analysed across arms within trials, in order to explore whether treatment-related difficulties varied according to the timing and sequence of a surgical technique. Parent reports of the child's treatment-related problems were also compared to recorded post-surgical medical complications (such as anaesthetic, airway, bleeding or other complications) using chi square analyses. Exact tests were used to calculate levels of significance. Since no disciplines were suggested in one of the five participating countries, an overall category (general treatment-related problems) was created in order to be able to compare the results across all countries. This variable included both the general and the specific comments regarding reactions to treatment.

RESULTS

Cleft Evaluation Profile

Mean scores and standard deviations, for the whole sample are presented in Table I. Parents were least satisfied with teeth and bite, followed by nose and speech. Parents of boys were more satisfied with their child's bite ($p < .05$), lip ($p < .01$), nose ($p < .001$), profile ($p < .01$), and total CEP score ($p < .05$), than parents of girls.

INSERT TABLE I ABOUT HERE

General satisfaction with treatment and appearance

The cut-off score of 32 on the total score for all items [8] showed that 88.7% ($n = 250$) of the parents were found to be satisfied, while 11.3% ($n = 32$) were dissatisfied. The number of parents scoring above the cut-off score was similar across all three trials and arms ($p > .05$).

Armwise within trials

There were few significant differences between arms within the three trials. Satisfaction with speech was the only significant variable in Trial 1, parents from Arm A, hard palate closure at 12 months being more satisfied than parents in Arm B, hard palate closure at 36 months ($p < .01$). In Trial 2, parents from Arm C were more satisfied with their child's bite than parents from Arm A ($p < .05$). There were no significant differences between Arm A and D in Trial 3.

Armwise across trials

Means and standard deviations for arms and trials are found in Table I. Post Hoc tests revealed that there were no differences between arms in satisfaction with speech, hearing, nose, or breathing. The main differences were found between Arm B and D, parent's from Arm B being more satisfied with teeth ($p < .05$), lip ($p < .05$), profile ($p < .05$), and total score ($p < .01$). Parents also reported more satisfaction with teeth in

Arm B than A ($p < .05$), and with bite compared to Arm D ($p < .05$). Parents from Arm C were more satisfied on the total score than parents from Arm D ($p < .05$).

The Scandleft Parent Questionnaire: Coping with treatment

A total of 85.1% ($n = 303$) of the parents responded to the question about treatment-related difficulties. Among those, 80.5% ($n = 244$) reported that the child had coped well with treatment. Parents reported minor and/or short-term problems as a result of cleft-related treatment experiences in 17.5% ($n = 53$) of the children, described as difficulties with sleep, eating, pain, and minor emotional reactions to hospital environments. The remaining 2.0% ($n = 6$) reported major or lasting problems, such as severe sleep problems, health related or behavioural changes, and/or anxiety levels significantly impacting on treatment and/or everyday life.

INSERT TABLE II ABOUT HERE

As can be seen in Table II, parents reported treatment related problems in approximately one third of the children within Trial 1, irrespective of Arm. There were more treatment related problems reported for Arm A than Arm B, C, and D across all trials. However, none of the differences across arms within the three trials were statistically significant ($p > .05$). When comparing parent reports with objective measures of post-surgical complications, analyses were non-significant (Range $\chi^2 = 0.08 - 4.10$, $p > .05$), irrespective of type of complication (anaesthetic, airway problems, bleeding, or other).

Speech therapy

A total of 61.0% ($n = 217$) of the parents provided specific information to this question, among whom 93.5% ($n = 203$) reported that the child had coped well with speech therapy and visits to the cleft center (Table II). Parents from Trial 2 and 3

reported less than 6% children with minor treatment-related problems after speech therapy, and none with major/lasting treatment-related problems. One third (31.3%; $n = 5$) of the parents from Trial 1, Arm B reported treatment-related problems which impacted on the child's response to speech therapy. However, none of the differences across arms and trials were statistically significant ($p > .05$).

Surgery

A total of 68.0% ($n = 242$) of the parents provided specific information to this question. As can be seen in Table II, 77.3% ($n = 187$) of these parents thought that their child had coped well with surgical treatment. While parents in Trial 2, Arm C (92.7%) and Trial 3, Arm D (89.8%) reported few treatment-related problems, approximately 20% of the parents from Arm A (Trial 2 and 3) reported difficulties after surgery. In Trial 1, more than half of the parents from Arm A (minor: 46.2%; major: 11.5%) and Arm B (minor: 46.4%; major: 7.1%) reported post-surgical problems in the child. Parent reports of treatment-related difficulties were not related to any objective measures of post-surgical complications (Range $\chi^2 = 0.01-3.79$, $p > .05$), irrespective of type of complication (anaesthetic, airway problems, bleeding, or other).

DISCUSSION

The present study explored parents' satisfaction with their child's treatment and appearance. According to the Cleft Evaluation Profile, the majority of parents were satisfied with their child's cleft-related features and appearance. Additionally, most parents indicated that the child had coped well with the burden of care. Some parents from Trial 1, however, reported a disturbingly high frequency of emotional and/or

behavioural problems after surgical treatment. Parent reported difficulties were not associated with objective records of post-surgical treatment complications.

Parents' satisfaction with cleft-related features and appearance

In general, parents were satisfied with cleft-related features and their child's appearance, as indicated by mean scores below or close to 3 on the CEP. Other studies based on the same measure have reached similar conclusions [6,8]. In general, parents reported least satisfaction with teeth and bite, followed by nose, speech and lip, very similarly to Noor and Musa [8]. This is not surprising, given that orthodontic treatment has not yet started when the child is 5 years old, and corrective surgery also will occur until late adolescence. Similarly, speech problems are known to be prevalent in younger children with a cleft (for a review see [18]), and most children in need of a surgical intervention because of velopharyngeal insufficiency had not yet undergone this surgery by the age of 5.

Analyses across trials and arms indicated few statistically significant differences.

When comparing parents' views regarding the timing and sequence of a surgical technique, parents in Arm B (later palate closure) were more satisfied than parents in Arm A and D with teeth and bite, while Trial 1 indicated that parents in Arm A (earlier palate closure) were more satisfied with speech than parents from Arm B. Further, Trial 2 pointed to parents from Arm C (soft and hard palate closure at 12 months) being more satisfied with bite than parents from Arm A (soft palate at 3 months).

Several studies have used the CEP [5,6,7,8,9,10] and overall comparisons indicate very similar results, and are also in line with the more recent Cleft Care UK study [19]. Most parents in the present study (88.7%) had scores indicating they were

satisfied with treatment and their child's appearance, compared to 75% in Noor and Musa's study [8]. Mean scores also indicated adequate satisfaction with their child's cleft-related features. Unfortunately, the CEP has been used differently across studies. As an example, some studies have restricted the number of items, while others have changed the direction of the scale, complicating or impeding comparisons between the studies. In addition, the above mentioned studies are based on data from older participants (age 10, adolescents, or young adults) rather than parents of younger children, and different cleft types.

Parent's satisfaction with their child's appearance may be helpful in considering differences between surgical protocols, however, there are limitations in these data. The CEP offered a snapshot of parental satisfaction with their child's appearance and other cleft-related features at 5 years. These perceptions cannot be considered purely as reflection of satisfaction with the outcomes of treatment as they are likely to have been affected by additional factors, including for example, the unconditional acceptance parents may feel for their child. One study demonstrated that inter-rater agreement between patients and parents tends to be moderate [11], and that professionals were more satisfied with treatment outcomes than parents [11]. Previous research has reported only weak associations between subjective and more "objective" measures of cleft-related features [14,20,21,22]. Hence, centers with lower parental satisfaction scores compared to other centers do not necessarily have poorer aesthetic outcomes. Differences could be related to social and cultural differences, or differing expectations regarding treatment. Further, an association has been reported between parental satisfaction with treatment outcomes and social interactions [11], and should be investigated in future studies.

Coping with treatment

Children with CLP are exposed to several medical procedures in the course of their first years of life, leading some researchers to ask whether they may be at risk for diffuse or procedure-specific anxiety [4,23,24]. Results have so far not been conclusive, and there is a need for longitudinal designs with large samples, including control groups from the normative population.

In the present study, the majority of parents (80.5%) reported that their child was coping well with treatment. When specifically asked about speech therapy and surgery, results indicated, as might be expected, that speech therapy, being less invasive and not associated with physical pain, was less challenging and anxiety-provoking for the child than hospitalisations and surgical treatment. A disturbing finding was the high frequency of treatment-related problems apparent in Trial 1. Analyses across trials revealed that while less than 6% of the children from Trial 2 and 3 (Arm A, C, D) had treatment-related problems with speech therapy, 31.3% of the children in Arm B, and 15.4% of those in Arm A, had treatment-related difficulties in Trial 1. This finding was even stronger for surgical treatment (Trial 1: 46.2-46.4%; Trial 2 and 3: Range 7.3-17.4%). These results have to be interpreted with caution, since the subsamples of parents reporting treatment-related problems within trials were small (see Table II). Nevertheless, an interesting observation is that parents reported fewer (however non-significant) treatment-related problems for the original surgical method in Trial 2 and 3. This finding could reflect a lack of felt competence in cases of a change of protocol, which possibly could affect parents' perception of their child's coping. However, this interpretation is questionable and the dataset does not allow for any further clarifications. The same applies for the high

frequency of parent reported difficulties in Trial 1. The results could suggest cultural and social differences between the countries included in the respective trials, such as differences in levels of openness about psychological adjustment and willingness to share or perceive difficulties, an interpretation that would have to be investigated by future studies.

The present study cannot answer questions about the reasons for the development of treatment-related problems in some of the children. Interestingly, when comparing recorded post-surgical complications (such as anaesthetic, airway, bleeding or other complications) with parents' perceptions, no association was found between the "objective" and "subjective" measures of treatment-related problems. Hence, post-surgical complications did not explain parents' perception of treatment-related difficulties in the child. Most probably, many different factors are involved in the development of treatment-related anxiety [4,25], such as quantity and intensity of treatment, previous anxiety-provoking experiences, the child's temperament, parenting style, and the availability and number of coping mechanisms [24]. Previous research has also pointed to the importance of the provision of information about upcoming procedures, risks, and possible post-operative consequences [26,27], in addition to parental concerns regarding aftercare following the child's operations [19], factors that seem to contribute to the way young patients and their parents cope with treatment. Another potential factor of importance is that only three of the nine cleft centres that participated in the current study had a clinical psychologist working within the team. Interestingly, two of the cleft centres within the trial reporting the highest parent-reported treatment difficulties, actually have a psychologist available within the team. An interesting question could therefore be whether parents could be more aware of potential post-surgical problems in their child, and hence more open

about them in the questionnaire. On the other hand, this effect was not found in the third centre having a psychologist within the team. In most cleft centres, parents were followed up by local health services if needed, with a potential significant variation in the level of available support.

Strengths and limitations

While studies on cleft lip and palate are mostly based on single centre studies, the major strength of the present dataset was its inclusion of nine centers and five countries. Additionally, the study was based on a randomised controlled trial, the most rigorous way of determining whether a cause-effect relationship exists between cleft treatment and outcomes. Further, all children were aged 5, providing an interesting cross-sectional picture of parent satisfaction with their child's cleft-related features and coping with treatment at one particular developmental stage. In addition, response rates were high (78-80%) and based on consecutive birth cohorts.

The study also has a number of limitations that should be taken into account. First, the Scandcleft Parent Questionnaire was administered retrospectively, and based on parent reports only, with the exception of recorded post-surgical complications. Further, some differences between teams could potentially have affected parental responses. One team sent the questionnaire to the parents by post before attendance to the 5-year-old assessment, while the other teams handed out the questionnaires face-to-face. Only three of the nine cleft centres had a clinical psychologist working within the team. However, parents completed the questionnaires while waiting for their appointments at the cleft centre, and an individual meeting with the team's psychologist was not part of routine care at age 5 years, probably reducing this potential difference in how data were collected. Second, there were several

methodological weaknesses in parent reports on treatment-related difficulties; parent reports did not include validated measures of anxiety levels and treatment-related difficulties. Further, the question about treatment related problems was open-ended, which probably reduced both reliability and validity. Forced responses with a selection of choices might have led to different results. On the other hand, this potential limitation also means that the prevalence of reported difficulties have probably not been overestimated, since no answers were suggested. Additionally, one of the countries had chosen not to specify whether potential complications were related to surgical treatment or speech therapy, complicating the comparisons across countries, trials and arms. Further, the number of missing data was high in relation to this second aim. Unfortunately, we do not have information about the reasons for missing data. Parents may have felt the questions were not relevant for their child, they may have found it difficult to respond, or they may have found it difficult to tell the team about their child's problems with coping. Conclusions based on the present findings should therefore be drawn with care. In addition, speech therapy could be local or within the cleft team, again weakening the interpretation of the results. Third, approximately 20% of the participating parents did not hand in the Scandcleft Parent Questionnaire, and differences between participants and non-participants cannot be ruled out. Last, the comparison of outcome measures across countries based on larger samples should be the attention of future research.

CONCLUSIONS

The findings from the present study provide a valuable insight into parental satisfaction with appearance and other cleft-related features in their 5 year old children, and their child's coping with treatment at one point in time. Few if any studies have explored parents' perceptions of treatment-related challenges and post-

surgical problems in children with a cleft. Parents reported high levels of satisfaction with cleft-related features and their child's appearance. Nevertheless, they also reported a number of challenges related to their child's treatment experiences. Treatment-related anxiety and difficulties were described and included behavioural and emotional changes in some children post-surgery. These children should be identified and offered appropriate support and help. The results thus contribute to highlight the importance for cleft clinicians of assessing potential post-surgical problems, in order to prevent the development of more severe problems that could impact on adjustment and further treatment.

References

- [1] Semb G, Enemark H, Friede H, Paulin G, Lilja J, Rautio J, Andersen M, Åbyholm F et al. Scandcleft randomised control trials of primary surgery for patients with unilateral complete cleft lip and palate. Planning and management. Submitted to J Plastic Surgery and Hand Surgery.
- [2] Fitzsimons KJ, Mukarram S, Copley LP, Deacon SA, van der Meulen JH. Centralisation of services for children with cleft lip or palate in England: A study of hospital episode statistics. BMC Health Services Research 2012;12:148.
- [3] Wehby GL, Cassell CH. The impact of orofacial clefts on quality of life and healthcare use and costs. Oral Dis 2010;16:3-10.
- [4] Hermes D, Matthes M, Saka B. Treatment anxiety in oral and maxillofacial surgery. Results of a German multi-centre trial. J Craniomaxillofac Surg 2007;35:316-21.
- [5] Thomas PT, Turner SR, Rumsey N, Dowell T, Sandy JR. Satisfaction with facial appearance among subjects affected by a cleft. Cleft Palate Craniofac J 1997;34:226-31.
- [6] Turner SR, Thomas PWN, Dowell T, Rumsey N, Sandy JR. Psychological outcomes amongst cleft patients and their families. Br J Plast Surg 1997;50:1-9.
- [7] Williams AC, Bearn D, Mildinhall S, Murphy T, Sell D, Shaw WC, Murray JJ, Sandy JR. Cleft lip and palate care in the United Kingdom: The Clinical Standards Advisory Group

(CSAG) Study. Part 2: Dentofacial outcomes and patient satisfaction. *Cleft Palate Craniofac J* 20013;8:24-9.

[8] Noor SN, Musa S. Assessment of patients' level of satisfaction with cleft treatment using the Cleft Evaluation Profile. *Cleft Palate Craniofac J* 2007;44:292-303.

[9] Munz SM, Edwards SP, Inglehart MR. Oral health-related quality of life, and satisfaction with treatment and treatment outcomes of adolescents/young adults with cleft lip/palate: an exploration. *Int J Oral Maxillofac Surg* 2011;40:790–96.

[10] Van Lierde KM, Dhaeseleer E, Luyten A, Van De Woestijne K, Vermeersch H, Roche N. Parent and child ratings of satisfaction with speech and facial appearance in Flemish pre-pubescent boys and girls with unilateral cleft lip and palate. *Int J Oral Maxillofac Surg* 2012; 41:192-99.

[11] Gkantidis N, Papamanou DA, Christou P, Topouzelis N. Aesthetic outcome of cleft lip and palate treatment. Perceptions of patients, families, and health professionals compared to the general public. *J Craniomaxillofac Surg* 2013;41:e105-10.

[12] Hunt O, Burden D, Hepper P, Johnston C. The psychosocial effects of cleft lip and palate: a systematic review. *Eur J Orthod* 2005;27:274–85.

[13] Rumsey N, Stock NM. Living with a cleft: psychological challenges, support and intervention. In: Berkowitz S, ed. *Cleft Lip and Palate: Diagnosis and Management*. 3rd ed. Berlin, Heidelberg: Springer- Verlag. 2013:907–17.

[14] Moss TP. The relationships between objective and subjective ratings of disfigurement severity, and psychological adjustment. *Body Image* 2005;2:151–59.

[15] Ong J, Clarke A, White P, Johnson M, Withey S, Butler PE. Does severity predict distress? The relationship between subjective and objective measures of appearance and psychological adjustment, during treatment for facial lipoatrophy. *Body Image* 2007;4:239-48.

[16] Rumsey N, Harcourt D. Visible difference amongst children and adolescents: Issues and interventions. *Dev Neurorehabil* 2007;10:113-23.

[17] Semb G, Brattström V, Mølsted K, Prah-Andersen B, Zuurbier P, Rumsey N, Shaw WC. The Eurocleft study: Intercenter study of treatment outcome in patients with complete cleft lip and palate. Part 4: Relationship among treatment outcome, patient/parent satisfaction, and the burden of care. *Cleft Palate Craniofac J* 2005;42:83-92.

[18] Kuehn DP, Moller KT. Speech and language issues in the cleft population: The state of the art. *Cleft Palate Craniofac J* 2000;37:348-1-35.

[19] Waylen A, Ness AR, Wills AK, Persson M, Rumsey N, Sandy JR. Cleft care UK study. Part 5: Child psychosocial outcomes and satisfaction with cleft services. *Orthod Craniofac Res* 2015;18:47-55.

- [20] Vegter F, Hage JJ. Lack of correlation between objective and subjective evaluation of residual stigmata in cleft patients. *Ann Plast Surg* 2001;46:625-29.
- [21] Marcusson A, Paulin G, Ostrup L. Facial appearance in adults who had cleft lip and palate treated in childhood. *Scand J Plast Reconstr Surg Hand Surg* 2002;36:16-23.
- [22] Byrne M, Chan JC, O'Broin E. Perceptions and satisfaction of aesthetic outcome following secondary cleft rhinoplasty: Evaluation by patients versus health professionals. *J Craniomaxillofac Surg* 2014;42:1062-70.
- [23] Whitehead TD, Tobiasen JM, Hiebert JM. Presurgical anxiety treated with cognitive behavioral therapy in a 13-year-old female with cleft lip and palate: a psychological case report. *Cleft Palate Craniofac J* 1996;33:258-61.
- [24] Vogels WE, Aartman IH, Veerkamp JS. Dental fear in children with a cleft lip and/or cleft palate. *Cleft Palate Craniofac J* 2011;48:736-40.
- [25] Ten Berge M, Veerkamp JS, Hoogstraten J. The etiology of childhood dental fear: the role of dental and conditioning experiences. *J Anxiety Disord* 2002;16:321-29.
- [26] Chuacharoen R, Ritthagol W, Hunsrisakhun J, Nilmanat K. Felt needs of parents who have a 0- to 3-month-old child with a cleft lip and palate. *Cleft Palate Craniofac J* 2009;46:252-57.
- [27] Nelson PA, Kirk SA. Parents' perspectives of cleft lip and/or palate services: A qualitative interview. *Cleft Palate Craniofac J* 2013;50:275-85.